

AN INVESTIGATION INTO THE PRIMARY AND SECONDARY EFFECTS
OF FUNCTIONAL BEHAVIOURAL ASSESSMENT BASED
INTERVENTIONS FOR SLEEP PROBLEMS IN SIX CHILDREN WITH
AUTISM

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Abstract

Sleep problems, including co-sleeping, are highly prevalent in children with autism spectrum disorder (ASD). Given the negative secondary effects associated with sleep problems, it is essential that effective treatments for sleep problems are identified. An evidence-based approach to treating challenging behaviours in children with ASD is the use of functional behavioural assessment (FBA). However, there is limited research into using this tool to formulate treatments for sleep problems in this population. This thesis is comprised of two studies. Study 1 was a single-case pilot study that included a 6-year-old boy with ASD, which investigated the impact of sleep interventions on his sleep outcomes and parents wellbeing. Study 2 was a single-case multiple baseline across participants design which included five 2-6 year old children with ASD. Children in both studies demonstrated multiple sleep problems, including co-sleeping. Study 2 built upon the experimental design and methodology of the pilot study, and explored the collateral effects of improved sleep outcomes on children's daytime functioning, ASD symptomatology, and parent's sleep and partner relationship quality. FBA was used to inform individualised and function-based multicomponent interventions for all children in both studies. One participant withdrew from the study before completing intervention, and another was still involved in intervention at the time of submission of this thesis. In response to treatment, parental presence during sleep onset was eliminated for all six children, and co-sleeping following a night waking was eliminated for all children who completed intervention. For the families who completed intervention, improvements were seen in other sleep outcomes, with reductions

in sleep onset latency and night wakings for all children. Results of the Depression Anxiety and Stress Scales (DASS-21), Pittsburgh Sleep Quality Index (PSQI), and Relationship Quality Index (RQI) demonstrated improvements in sleep quality for all parents, some improvements in levels of depression, and mixed outcomes for relationship quality. Results on the Child Behaviour Checklist (CBCL) and Gilliam Autism Rating Scale (GARS-3) suggested improvements in the children's externalising behaviours, specifically attention, aggression and ADHD characteristics, as well as improvements in their overall ASD symptomatology, in particular their restricted/repetitive behaviours. The findings have important implications for the use of FBA to inform treatments for sleep problems that include co-sleeping, in children with ASD. Findings add to the scarcity of literature experimentally investigating pre- and post- measures of secondary problems associated with sleep disturbances.

Chapter 1

Autism Spectrum Disorder and Sleep

Sleep problems are highly prevalent among children with Autism Spectrum Disorder (ASD), and can have a negative impact upon children's daytime functioning (Goldberg & Keller, 2007; Teti, Shimizu, Crosby, & Kim, 2016), as well as family functioning and parental wellbeing (Teti et al., 2016). Unwanted co-sleeping, i.e. a child sleeping in close proximity to an adult, is a particular sleep difficulty that is highly prevalent in children with ASD (Liu, Hubbard, Fabes, & Adam, 2006). To date, the majority of research has focused on behavioural interventions for sleep problems in typically developing children, and less is known about the effectiveness of behavioural interventions in treating sleep problems in children with ASD (Deliens, Leproult, Schmitz, Destrebecqz, & Peigneux, 2015; Richdale & Wiggs, 2005; Turner & Johnson, 2012). There are also very few studies to have focused on treatment for co-sleeping. Functional Behavioural Assessment (FBA) is emerging as a valuable means to understand the nature of the child's sleep problems, allowing for the development of an individualised intervention treatment that targets the factors maintaining the child's difficulties, therefore increasing the likelihood of a positive outcome (Jin, Hanley, & Beaulieu, 2013).

The aim of the current study is to add to the current literature relating to sleep interventions in children with ASD, by examining the effectiveness of FBA to inform interventions for sleep problems that include co-sleeping, and to evaluate the effect of these sleep interventions on sleep-related outcomes.

In this chapter, ASD and associated challenging behaviours are described. Sleep patterns and problems in children with ASD are explored, with a focus on behavioural insomnias, including co-sleeping. Causes of sleep problems in children with ASD are discussed. Common interventions for sleep disorders, and the FBA process is introduced.

Autistic Spectrum Disorder

Definition. Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder that is characterised by persistent deficits in social communication and interaction and restricted and repetitive patterns of behaviour (American Psychiatric Association (APA), 2013). Aspects of social communication and interaction that may be impaired include nonverbal communicative behaviours, social-emotional reciprocity, and relationships (APA, 2013). This can manifest in a variety of ways, for example abnormalities in eye contact and body language, a failure to initiate or respond to social interactions, and difficulties making friends (APA, 2013). Behavioural symptoms can include, but are not limited to, repetitive motor movements, an insistence on sameness, intense and fixated interests, and hypo- or hyper-reactivity to sensory aspects of the environment (APA, 2013). These symptoms are present from early childhood, are not explained by an intellectual disability, and cause a clinically significant impairment in the individual's adaptive behaviour (APA, 2013). Efforts to describe individuals with characteristics of ASD have been found in the literature as early as 1944 (Elsabbagh et al., 2012).

While there are fundamental characteristics common to all individuals with ASD, there is great variability in the manifestation and severity of the symptoms (APA, 2013). Intervention, compensation, and level of support may mask the severity of the disorder. Each child's developmental level, the presence of comorbid conditions (Johnson, Burkett, Reinhold, & Bultas, 2016), and chronological age may also add to the heterogeneity in the clinical presentation of symptoms and the severity of impairment amongst this population (APA, 2013). As a result, ASD is a collective term that encompasses a range of symptom presentations (Stores & Wiggs, 1998; Van Wijngaarder-Cremers et al., 2014).

Prevalence. Approximately one in every 88 children in New Zealand has a diagnosis of ASD. This equates to approximately 50,000 children (Autism New Zealand, 2014). Recent data from the US estimates that approximately one in every 68 children has ASD (Christensen, 2016), and that the ratio of males to females diagnosed with ASD is approximately 4:1 (Johnson et al., 2016; Myers & Challman, 2011; Van Wijngaarden-Cremers et al., 2014). The prevalence of ASD is thought to have increased nearly 15 fold since the 1980's (Myers & Challman, 2011). Whether or not the increase reflects a true indication of rates of the disorder is a topic of debate (Myers & Challman, 2011; Saracino, Noseworthy, Steiman, Reisinger & Fombonne, 2010; Williams et al., 2014). An increase in prevalence may represent a true increase in the incidence of ASD's, or it may be due to heightened awareness of the disorder in both the lay and professional public; an increasing need to obtain autism diagnoses to ensure access to services; the development of screening and diagnostic tools to allow professionals to better identify individuals with ASD across a wide range of ages

and intellectual abilities; or broadening of the diagnostic criteria over time (Elsabbagh et al., 2012; Myers & Challman, 2011; Saracino et al., 2010; Williams et al., 2014). The high occurrence of this disorder makes individuals with ASD one of the highest priority populations for clinical research and treatment development (Cohen, Conduit, Lockley, Rajaratnam & Cornish, 2014).

Etiology of ASD. The cause of ASD is likely to involve multiple pathways and mechanisms, influenced by a complex combination of genetic, environmental and epigenetic factors (Fakhoury, 2015; Williams et al., 2014). Due to the complexity and heterogeneity of ASD, no single factor has yet been identified that conclusively explains the likelihood of getting the disorder (Fakhoury, 2015).

Genetic factors. Current thinking is that genetic factors contribute toward ASD, with heritability estimates as high as 90% (APA, 2013; Fakhoury, 2015). Currently, approximately 15% of cases of ASD appear to be associated with identifiable singular genetic mutations (APA, 2013; Myers & Challman, 2011), with different variants associated with ASD in different families (APA, 2013). The remaining 85% of cases appear to be polygenic, with mutations in numerous genetic loci making minute contributions (APA, 2013). Studies mapping chromosomes have identified several gene abnormalities that are associated with ASD, including mutations in brain-expressed genes such as GABA receptor subunit genes, serotonergic genes, dopaminergic genes, and those that impact on the development of mirror neurons and neuroligins (Fakhoury, 2015; Miano et al., 2007). Combinations of genetic factors help to explain the impairments and ranges of severity of each diagnostic component seen in individuals with ASD (Myers & Challman, 2011).

Environmental factors. Exposure to environmental stimuli can have everlasting effects on the developing brain, and can influence neurological processes such as cell differentiation, synaptogenesis and axon myelination (Fakhoury, 2015). A combination of environmental factors are likely to be required to have any significant influence on the predisposition of ASD (Fakhoury, 2015). Environmental factors that are known to increase the risk of ASD include aspects of the prenatal environment, such as chronic in-utero exposure to tobacco, alcohol and recreational drugs, maternal deficiencies in essential nutrients and fatty acids, and medications, especially those used to treat bipolar disorder and depression (APA, 2013; Fakhoury, 2015; Myers & Challman, 2011). Perinatal factors such as prematurity, low birth weight, and intrapartum hypoxia may also be associated with ASD (APA, 2013; Myers & Challman, 2011). Other factors such as exposure to air pollutants, poor socio-economic status, low maternal education level, and advanced paternal age have also been associated with ASD development (APA, 2013; Fakhoury, 2015).

Epigenetic factors. Like most disorders, ASD is most likely a result of very complex interactions between an individual's environment and their genetic profile (Fakhoury, 2015). Epigenetic changes due to environmental factors directly acting on susceptible genes could lead to structural changes in brain anatomy that are consistent with the abnormal cognition and social functions seen in individuals with ASD (Fakhoury, 2015). For example, Mazina et al. (2015) found that children with ASD who had ASD-associated copy number variants (CNV's) were more susceptible to the negative impacts of being prenatally exposed to maternal infection. The autistic children with the CNV's that were exposed to maternal infection had more severe core ASD symptoms

than other children with ASD who did not have the CNV's but were exposed to maternal infection, or autistic children who did have CNV's and were not exposed to maternal infection (Mazina et al., 2015).

As well as different causes resulting in wide variations in behaviours, it is also possible that many different causal pathways can lead to the same behaviours (Von Bertalanffy, 1967; Williams et al., 2014). Worldwide research is currently being undertaken, with the mammoth task of determining the factors that contribute to the etiology of ASD (Williams et al., 2014).

Challenging Behaviours in Autism Spectrum Disorders

Studies have suggested that 13-30% of typically developing young children display challenging behaviours that warrant intervention (Horner, Carr, Strain, Todd, & Reed, 2002). Estimates of children with ASD exhibiting some form of challenging behaviour are as high as 92% (Murphy, Healer, & Leader, 2009). These rates of challenging behaviour are significantly higher for children with ASD than for children with typical development (Baghdadli, Pascal, Grisi, & Aussilloux, 2003; Eisenhower, Baker, & Blacher, 2005; Hanley, Jin, Vanselow, & Hanratty, 2014; Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; Lecavalier, Leone, & Wiltz, 2006; McStay, Dissanayake, Scheeren, Koot, & Begeer, 2013; Murphy et al., 2005; Murphy et al., 2009). Common challenging behaviours that are part of the diagnostic criteria for children with ASD included extreme distress to small changes, eating difficulties, difficulties transitioning between activities, and negative reactions to particular sounds or textures (APA, 2013; Murphy et al., 2005). Challenging behaviours that are not required for a

diagnosis of ASD, but which covary with ASD at a high rate, include sleep difficulties, irritability, tantrums, aggression, self-injury, property destruction, hyperactivity, impulsivity, inattention, mood lability, pica, and inappropriate sexual expression (American Psychiatric Association, 2013; Dominick, Lainhart, Tager-Flusberg, & Folstein, 2007; Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Gray, 2002; Hanley et al., 2014; Horner et al., 2002; McStay et al., 2013; Myers & Challman, 2011; Myers & Johnson, 2007). Individuals with ASD are also often diagnosed with co-existing psychiatric disorders, such as depression, anxiety, attention-deficit hyperactivity disorder, and obsessive-compulsive disorder (APA, 2013; Kim et al., 2000; Myers & Challman, 2011).

In children with ASD, heightened behavioural problems are typically present before 3 years of age (Dominick et al., 2007; Eisenhower et al., 2005; Hanley et al., 2014). Although some challenging behaviours do appear to improve with age, there is considerable chronicity in challenging behaviours over time for those autistic individuals with the most pervasive problem behaviours (Eisenhower et al., 2005; Murphy et al., 2005).

These challenging behaviours are more likely than any other factor (including severity of the disorder) to impact on the physical and mental health, as well as the quality of life, of the individual (Matson & Nebel-Schwalm, 2007; Murphy et al., 2009; Myers & Johnson, 2007), and are one of the most significant stressors for their family and carers (Eisenhower et al., 2005; Gray, 2002; Hastings & Brown, 2002). Families of children with developmental disorders are at risk of more negative psychological outcomes than families with typical developing children (Gray, 2002; Lecavalier et al., 2006; McStay et al., 2013), and more negative effects are found on the health, wellbeing, and social experiences

of families with a child with autism than other developmental disabilities (Eisenhower et al., 2005; Lecavalier et al., 2006; McStay et al., 2013). Regardless of the autistic child's behaviour difficulties, their parents are more likely to have significant levels of depression, anxiety, anger, physical health problems and career problems (Gray, 2002). In addition, parents of children with ASD report higher levels of negative relationships with extended families, especially grandparents, than other developmental difficulties (Gray, 2002), resulting in lower levels of support and fewer resources to deal with their situation. Challenging behaviours that are not part of the criteria for an ASD diagnosis per se have been found to predict even greater stress in parents of children with autism and appear to be more strongly associated with parent stress than any other child or caregiver characteristic (Lecavalier et al., 2006; Gray, 2002; McStay et al., 2013). Longitudinal studies have found continuity in the negative impacts of these behaviours on families (Eisenhower, 2005; Gray, 2002).

One of the most burdensome and common challenging behaviours reported by parents of children with ASD is sleep difficulty (Brown et al., 2014; Cohen et al., 2014; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Polimeni, Richdale, & Francis, 2005). It is likely that sleep problems maintain and exacerbate daytime behaviour problems in children with ASD (Vriend, 2011). Treating disordered sleep in children with ASD represents a potential avenue to improve daytime behaviour and family functioning in this population (Meltzer, 2008).

Sleep in children with Autism Spectrum Disorder

The function of sleep. Children spend more time in sleep than they do in any other activity (Arbelle & Ben-Zion, 2001; Lushington, Pamula, Martin, & Kennedy, 2013; Wiggs, 2007). The function of sleep remains largely unknown, however it is recognised that sleep is a developmental process that co-occurs, co-regulates and is causally linked with other developmental processes, including behaviour and emotion regulation, learning and attention, social interactions, memory consolidation, energy conservation, brain growth, and physical growth (Brown, Kuo, Phillips, Berry, & Tan, 2013; Deliens et al., 2015; Richdale, 2013; Staples, 2013; Stores & Wiggs, 1998; Turner & Johnson, 2012).

In typically developing children, reduced sleep quantity can increase the likelihood of internalising behaviour problems such as anxiety and depression, as well as externalising behaviour problems such as aggression and hyperactivity (Bagley & El-Sheikh, 2013; Lushington et al., 2013). Interactions have also been found between poor sleep quality and poor academic performance (Ahrberg, Dresler, Niedermaier, Steiger, & Genzel, 2012; Reale, Guarnera, & Mazzone, 2013; Schmidt & Van der Linden, 2015), as well as poorer health outcomes, including an increased risk of diabetes and obesity (Brown et al., 2013). Given the importance of sleep on daily functioning, the consequences of disrupted sleep in individuals with ASD and their families are potentially serious (Cohen et al., 2014; Krakowiak et al., 2008).

Prevalence of sleep problems. Sleep problems are a common and serious comorbid condition for individuals with ASD (Cortessi, Giannotti,

Ivanenko, & Johnson, 2010; Richdale, 2013; Wiggs & Stores, 2004). Estimates of sleep difficulties in children with ASD are high, ranging from 33-83% (Goldman et al., 2011; Kotagal & Broomall, 2012; Mannion, Leader & Healy, 2013; Miano et al., 2007; Park et al., 2012; Richdale & Schreck, 2009; Rzepecka, McKenzie, McClure & Murphy, 2011; Singh & Zimmerman, 2015) compared to 15-35% for children with typical development (Brown et al., 2013; Krakowiak, 2008; Richdale & Schreck, 2009; Singh & Zimmerman, 2015). Attaining an accurate estimate of the prevalence of sleep disorders in children with ASD is difficult, as the children themselves often do not complain about this problem (Picchioni, Reith, Nadel, & Smith, 2014;). They are rarely screened for and recognised by physicians (Meltzer, Johnson, Crosette, Ramos, & Mindell, 2010; Richdale & Wiggs, 2005), and caregivers are likely to only report disturbed sleep if they recognise this as a problem (Richdale & Wiggs, 2005; Weiskop, Richdale, & Matthews, 2005). Sleep difficulties are frequently not reported by parents as they often view such problems as a long-standing consequence of their child's symptomatology that is not resolvable, or they are more focused on managing other more debilitating and obvious challenges (Bartlet & Beaumont, 1998; Picchioni et al., 2014; Richdale & Schreck, 2009; Richdale & Wiggs, 2005; Robinson & Richdale, 2004). As a result, prevalence rates are potentially even higher than reported.

Course of sleep problems. Without intervention, sleep problems are found to be more pervasive and have lower rates of remission in children with ASD, when compared to typically developing children (Deliens et al., 2015; Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; Kodak & Piazza, 2008; May,

Cornish, Conduit, Rajaratnam, & Rinehart, 2015; Murphy et al., 2005; Richdale, 2013; Richdale & Schreck, 2009; Papadoulus et al., 2015; Siversten, Posserud, Gillberg, Lundervold, & Hysing, 2012). If untreated, childhood sleep problems are likely to persist into adulthood (Deliens et al., 2015; Didden et al., 2002; Dominick et al., 2007; Miano & Ferri, 2010; Richdale, 2013), and may actually increase and change in nature (Goldman, Richdale, Clemons, & Malow, 2012; Hodge et al., 2014; Sivertsen et al., 2012). As sleep problems are less likely to self-resolve through maturation alone in individuals with ASD, early intervention is essential for these children and families to avoid enduring sleep problems (Durand & Christodulu, 2004; Hodge et al., 2014).

Categories of sleep disturbance. According to Krakowiak et al. (2008), there are three major categories of sleep disturbance: behavioural insomnias, parasomnias, and secondary sleep disorders. Behavioural insomnias refer to maladaptive sleeping patterns that occur while the individual is awake, including difficulties falling asleep, problems maintaining sleep, and/or early morning awakenings (Krakowiak et al., 2008; Dahl, 1995; Roane & Taylor, 2013). Parasomnias include unusual or undesirable behaviours during sleep which intrude on the sleep, such as night terrors, nightmares, sleepwalking, sleeptalking, repetitive rhythmic behaviours, nocturnal seizures and enuresis (Krakowiak et al., 2008; Dahl, 1995). The final category is sleep disorders that are secondary to a physical illness or a psychological disorder. Patterns of behavioural insomnias are the most frequently reported type of sleep disturbance for children with ASD (Buckley et al., 2010; Cortesi, et al., 2010; Deliens et al., 2015; Didden & Sigafos, 2001; Goldman, et al., 2012; Hodge et al.,

2014; Miano et al., 2007; Polimeni et al., 2005; Richdale, 2013; Richdale & Schreck, 2009; Thirumalai, Shubin, & Robinson, 2002; Souders et al., 2009; Wiggs & Stores, 2004), and are therefore the focus of this report.

Behavioural insomnias in children with Autism Spectrum Disorder.

Research shows that sleep problems differ in intensity, duration and frequency for children with ASD compared to typically developing children and children with other developmental disabilities (Hodge et al., 2014; Polimeni et al., 2005; Richdale & Schreck, 2009). Commonly reported sleep problems in children with ASD are those associated with settling and sleep onset (increased sleep onset latency) and sleep maintenance (decreased sleep duration, decreased sleep continuity, and early wakings) (Deliens, et al., 2015; Cortesi, et al., 2010; Krakowiak, et al., 2008; Miano & Ferri, 2010; Miano et al., 2007; Reed et al., 2009; Richdale, 2013; Richdale & Schreck, 2009; Singh & Zimmerman, 2015; Thirumalai et al., 2002; Vriend et al., 2011; Wiggs & Stores, 2004). Unwanted co-sleeping is also highly prevalent in this population, and often occurs as a means to aid sleep onset and maintenance (Goldberg & Keller, 2007).

At bedtime, children with ASD are less likely to be sleepy, and are more likely to be noncompliant, have difficult and challenging bedtime behaviours, have nonfunctional and challenging bedtime routines or rituals, and require fluids or medications to fall sleep, all which contribute to a longer sleep onset latency (Miano et al., 2007; Richdale, 2013). They are more likely than typically developing children to have difficulties falling asleep again after wakings during the night (Miano et al, 2007; Richdale, 2013). Sleep onset problems are more common than sleep maintenance problems (Singh & Zimmerman, 2015).

However, sleep problems frequently coexist (Liu, et al., 2006; Spruyt & Curfs, 2015). For example, a child who has difficulty initiating sleep when first put down to sleep for the night is also likely to have difficulties resettling when they wake during the night (Wiggs & France, 2000).

Co-sleeping. Co-sleeping is a bedtime behaviour that can create problems for many families with and without ASD. Co-sleeping is defined as “... the presence of at least one... adult caregiver who sleeps within close enough proximity of the infant to permit the exchange of at least two sensory stimuli (touch, smell, movement, sight, and/or sound)” (McKenna & Volpe, 2007, p.1). Bed sharing, when an infant or child sleeps in the same bed as one or both parents, is considered a subtype of co-sleeping (Burnham, 2013; Goldberg & Keller, 2007), but given it is the most common definition of co-sleeping in lay terms, it is how co-sleeping will be defined in this thesis. An important distinction is made between intentional co-sleepers and reactive co-sleepers. (Goldberg & Keller, 2007; Keller & Goldberg, 2004; Ramos, Youngclarke, & Anderson, 2007). Intentional co-sleepers are families who consciously choose to co-sleep from early infancy onwards and place importance on this arrangement (Goldberg & Keller, 2007). Reactive co-sleeping refers to children who co-sleep due to difficulties sleeping alone even though their parents prefer separate sleeping arrangements, and usually occurs after the child is 1 years old (Goldberg & Keller, 2007; Keller & Goldberg, 2004; Ramos et al., 2007). Children who sleep with their parents for only part of the night are more likely to be reactive co-sleepers, demonstrating the parents desire for their child to sleep alone (Goldberg & Keller, 2007). In these families, children either fall asleep in

the parents bed and get transferred to their own bed during the night, the parents lie in the child's bed until they fall asleep and then leave, or the child starts sleep in their own room, often with parental assistance, but upon night wakings are unable to self-soothe and require co-sleeping to return to sleep (Goldberg & Keller, 2007). It is the latter that often causes the most distress for parents, as their sleep is also disrupted (Ward, 2015). When well rested, these parents often intend to return their children to their own beds, but in the middle of the night when exhausted themselves, and dealing with an incessantly difficult child, are inclined to allow their child to lie with them (Ward, 2015).

In the US, 5% of typically developing children aged 2 – 18 years old sleep with their parents, but this increases to 16% with children with ASD in this age group (Liu et al., 2006). For this age group, most co-sleeping children are reactive co-sleepers, compared with the majority of infants being intentional co-sleepers (Hayes, Fukumizu, Troese, Sallinen, & Gilles, 2007; Ramos et al., 2007).

Requiring parental assistance to sleep is a persistent problem (Gaylor, Burnham, Goodlin-Jones, and Anders, 2005; Hayes et al., 2007). A longitudinal study by Gaylor et al. (2005) found 33% of typically developing children required parental intervention to reinitiate sleep following a night waking at 6-12 months of age. These children were more likely than self-soothers to be reactive co-sleepers at 2 and 4 years of age (Gaylor et al., 2005).

Literature pertaining to infants suggest that reactive co-sleeping is predicted by a child's sleep onset difficulties, frequent night wakings, and high levels of sleep anxiety (Cortessi, Giannotti, Sebastiani, Vagnono, & Mariono, 2008). Although it is unclear as to how this relates to older children or children with developmental disabilities, reactive co-sleeping is likely to be correlated

with the same difficulties and maintained for similar reasons. Co-sleeping is an effective tool for decreasing bedtime resistance, sleep onset latency, and duration of night wakings, especially in anxious children who have not learnt to self-soothe (Richdale, 2013). Out of desperation or convenience, parents with these difficulties often let their children co-sleep (Keller & Goldberg, 2004). Due to the proximity of the child, parents are more aware of the small noises and movements that accompany a typical night waking (Goldberg & Keller, 2007; Keller & Goldberg, 2004; Ramos et al., 2007). It is possible that because parents of reactive co-sleepers are more likely to perceive night wakings as a problem, they are more likely to give attention to the child following a night waking. This however, is likely to maintain co-sleeping, as the child does not learn to self-soothe.

For families co-sleeping with a child with ASD, additional factors may account for co-sleeping (Liu et al., 2006). Parents may believe that due to their child's disabilities, they require more care and attention during the night (Liu et al., 2006). Also, the child's behaviours, for example difficulties with making transitions, extreme reactions to small changes, and sensory seeking behaviours, may contribute to them finding it more challenging to cease co-sleeping (Liu et al., 2006). Co-sleeping in families with children with ASD is therefore more likely to be a result of reactive co-sleeping than intentional co-sleeping.

Co-sleeping can have negative impacts on the child's sleep behaviours (Cortesi et al., 2004; Liu, Liu & Wang, 2003). In infants at least, children who co-sleep are more likely to have more daytime sleepiness and sleep anxiety, later bedtimes, and significantly shorter total sleep durations than children who do not co-sleep (Liu et al., 2003). In addition, they have more bedtime resistance,

and more behavioural and emotional problems (Cortesi et al., 2004; Liu, et al., 2003).

Reactive co-sleeping can have a negative impact on the families' wellbeing (Goldberg & Keller, 2007; Teti et al., 2016). In comparison to families with independent sleepers, it is correlated with more sleep disruption to the mother (Goldberg & Keller, 2007; Teti et al., 2016), as well as more marital distress, and lower emotional availability to their child at night time (Teti et al., 2016). In addition, reactive co-sleeping can be a marker of heightened family distress (Teti et al., 2016).

Sleep architecture in children with Autism Spectrum Disorder.

Studies have found that sleep architecture, which describes the structure and pattern of sleep in the brain, differs in children with ASD compared to typically developing children or children with other developmental disabilities (Arbelle & Ben-Zion, 2001; Buckley et al., 2010; Cortesi, et al., 2010; Elia et al., 2000; Miano et al., 2007; Richdale, 2013; Richdale & Schreck, 2009; Thirumalai, Shubin, & Robinson, 2002; Wiggs & Stores, 2004).

Research using polysomnographs (PSG), the gold standard for measuring sleep architecture, has found the presence of disrupted sleep architecture in children with ASD (Arbelle & Ben-Zion, 2001; Buckley et al., 2010; Elia et al., 2000; Miano et al., 2007; Richdale, 2013; Richdale & Schreck, 2009; Thirumalai et al., 2002). These studies have found that in comparison to typically developing children or children with other developmental difficulties, children with ASD are likely to spend less time in bed, have less total sleep time, increased sleep onset latency, earlier wake times and an increased number of night wakings (Buckley

et al., 2010; Elia et al., 2000; Miano et al., 2007; Richdale & Schreck, 2009).

Differences have also been discovered within the different stages of sleep.

Significant differences are found in REM sleep, and include shorter REM sleep latency, immature organization of eye movements with periods not becoming longer during the night, decreased eye movement density, lower percentage of REM sleep, absence of skeletal muscle atonia which normally prevents the acting out of dreams, and lack of dream reports when woken during this stage (Arbelle & Ben-Zion, 2001; Buckley et al., 2010; Elia et al., 2000; Richdale, 2013; Richdale & Schreck, 2009; Thirumalai et al., 2002). During non-REM sleep, children with ASD are more likely to have less slow wave sleep (SWS), lower cyclic alternating patterns during SWS with less A1 subtypes, an increased amount of stage 1 sleep, and a higher percentage of stage 3 sleep (Buckley et al., 2010; Elia et al., 2000; Richdale, 2013; Richdale & Schreck, 2009). Interestingly, these unusual sleep patterns are fairly stable across autistic individuals, regardless of reports of sleeplessness or not (Wiggs & Stores, 2004).

How altered sleep architecture relates to problematic sleep and ASD symptomology and etiology still remains to be determined (Richdale, 2013). However, it has been hypothesised that disrupted sleep architecture may be a consequence of a dysfunction in neurological processes (Buckley et al., 2010; Elia et al., 2000; Kotagal & Broomall, 2012; Miano et al., 2007). This research is still very much in its infancy and is a subject of ongoing research (Buckley, 2010).

Cause of sleep problems. The cause of sleep problems in children with ASD is not well understood (Gringas et al., 2014; Richdale, 1999). Factors contributing to sleep problems in children with ASD may be different to those

that cause sleep problems in typically developing children (Hodge et al., 2014; Richdale & Schreck, 2009). There are several theories regarding the etiology of sleep disturbances in children with ASD. It is likely, however, that since sleep requires a complex relationship between physiology, habits and behaviours, and social forces (Brown et al., 2013), sleep disturbances are the consequence of complex interactions between multiple factors, that may vary from child to child (Cortesi et al., 2010; Deliens et al., 2015; Goldman et al., 2012; Kotagal & Broomall, 2012; Krakowiak et al., 2008; Miano & Ferri, 2010; Papadopoulus et al., 2015; Reed et al., 2009; Richdale, 2013; Richdale & Schreck, 2009; Singh & Zimmerman, 2015). Richdale & Schreck (2009) proposed that the cause of sleep problems in this population be regarded from a biopsychosocial viewpoint.

Biological factors. Biological or genetic abnormalities observed in individuals with ASD alter brain architecture or biochemistry (Richdale, 2013). As described earlier, sleep architecture is different in individuals with ASD, but how it is related to problematic sleep is yet to be definitively determined. One mechanism may be through the irregular expression of several neurotransmitters, such as melatonin, serotonin, and gamma-amino butyric acid (GABA) that have been discovered in autistic individuals (Richdale, 2013). These neurotransmitters play important roles in the development and maintenance of sleep-wake cycles, for example through signaling sleep onset, synchronising circadian rhythms, and regulating sleep (Cortesi et al., 2010; Deliens, 2015; Richdale, 2013; Richdale & Schreck, 2009; Singh & Zimmerman, 2015; Stores & Wiggs, 1998).

Like other children, co-occurring medical conditions, such as allergies, asthma, and gastrointestinal problems, can also increase the likelihood of sleep

problems (Kotagal & Broomall, 2012; Richdale, 2013; Singh & Zimmerman, 2015). Patterns of behaviours that are established during periods of illness become normalised and incorporated in to the expected sleep routine (Didden et al., 2002). However, resolving sleep problems following the resolution of medical problems may be more difficult for children with ASD, who have greater difficulty reversing habits, routines or expectations that may have been set during this time (Didden et al., 2002; Richdale, 2013). In addition, approximately 20-40% of children with ASD also have epilepsy, which may be associated with sleep problems (Richdale, 2013). Medications to treat epileptic seizures can also disrupt sleep (Singh & Zimmerman, 2015).

Recent studies have also suggested that insomnia can result from nutritional deficiencies (Singh & Zimmerman, 2015). 50-70% of children with ASD have feeding behaviour difficulties such as food phobias, strong food preferences, and atypical mealtime rituals and behaviours, resulting in restricted diets and in increased risk of malnutrition (Kodak & Piazza, 2008; Singh & Zimmerman, 2015).

Psychological factors. Psychological and behavioural problems related to core and associated features of this disorder can also affect sleep (Malow et al., 2014). Communication and social difficulties can result in impaired awareness of social and environmental cues that are used to synchronise circadian rhythms, such as the natural light-dark cycle, sound, and understanding directions about falling asleep (Deliens et al., 2015; Kotagal & Broomall, 2012; Malow et al., 2014; Miano & Ferri, 2010; Richdale, 1999; Singh & Zimmerman, 2015; Stores, 1992). In addition, communication deficits can make it hard for these individuals to

express any pains or discomforts that may be hindering their sleep (Reed et al., 2009; Singh & Zimmerman, 2015).

Nonfunctional routines and unusual sleep rituals may lead to bedtime resistance and settling difficulties, especially when the conditions required for these routines are not met (Richdale, 1999; Richdale & Schreck, 2009). Given that individuals with ASD often have difficulties breaking routines, they are at an increased risk of not being able to fall back asleep following a spontaneous waking, when conditions they required to fall asleep with initially are no longer present (Deliens et al., 2015, Reed et al., 2009). Similarly, minor changes to a bedtime routine potentially result in prolonged sleep onset (Deliens et al., 2015).

Abnormalities in sensory sensitivity can impede sleep in these children (Cortesi et al., 2010; Deliens et al., 2015; Singh & Zimmerman, 2015). Children under-responsive to stimuli may miss factors that cue sleep, such as the natural light-dark cycle (Cortesi et al., 2010; Deliens, 2015). Being over-responsive to stimuli may also create sleeping difficulties, for example a child who is oversensitive to tastes or textures may be more anxious at bedtime when they need to brush their teeth, and hence have difficulty falling asleep when in that state (Deliens, 2015). It has also been suggested that children with ASD can be either hyper- or hypoaroused, and that they actually may require less sleep as a result (Deliens, 2015; Richdale, 1999).

Anxiety, depression and ADHD are associated with sleep problems in typically developing children (Richdale, 2013). Given these comorbid conditions are highly prevalent in the ASD population, they may also precipitate or maintain sleep problems in these individuals (Cortesi et al., 2010; Deliens et al., 2015; Richdale, 2013; Richdale & Schreck, 2009; Singh & Zimmerman, 2015).

Correlations have been found between autistic children's daytime behaviour and sleep patterns, with children who engage in disruptive and difficult behaviours, especially self-injury, aggression, mood swing, and compulsive behaviour being more likely to also be poor sleepers (Goldman et al., 2011; Richdale & Schreck, 2009). Most common sleep problems seen in autistic children, particularly bedtime resistance and failing to return to sleep after a spontaneous night waking, can be conceptualised as a continuation of the challenging behaviours themselves (Richdale & Schreck, 2009).

Unfortunately, all of the aforementioned factors that interfere with sleep can trap the individual in a feedback loop, whereby these conditions can worsen the sleep problem, and are in turn worsened themselves (Singh & Zimmerman, 2015).

Social factors. The child's social environment may impact sleep (Richdale, 2013; Richdale & Schreck, 2009; Cortesi et al., 2010; Richdale & Wiggs, 2005). Parents of children with ASD are at elevated risk of suffering from stress and depression, marital problems, and anxiety about their child, compared with others and report poorer sleep quality and quantity (Lopez-Wagner, Hoffman, Sweeney, Hodge, & Gilliam, 2008; Meltzer, 2008; Richdale, 2013; Richdale & Wiggs, 2005). Cortesi et al. (2010) found that parent sleep alone is able to predict the sleep problem severity of their child, however this is likely a bidirectional phenomenon. Richdale (2013) suggested that these parents are struggling with a multitude of challenges, which make it difficult to implement consistent, disciplined strategies conducive to creating good sleep patterns for their children.

The complex interaction between these factors must be considered when investigating the cause and treatment of sleep difficulties in children with ASD (Richdale & Schreck, 2009).

Behavioural model of sleep disturbance. While there are various underlying causal mechanisms, sleep problems can result from children having not learnt appropriate ways of getting to and staying asleep. A behavioural model based on operant behaviour theory can be used to explain sleep problems of insomnia (Didden et al., 2002).

Operant behaviour theory stipulates that what occurs before and after a behaviour impacts on the likelihood of that behaviour reoccurring in similar contexts or environments (Blampied, 2013; Skinner, 1969). The reoccurrence of behaviours are contingent upon the interrelationship between the antecedents that precede a behaviour, serving as discriminative stimuli that signal a behaviour to occur (A), the behaviour/response itself (B), and the consequences that directly follow the behaviour (C) (Skinner, 1969). Consequences can be either reinforcing or punishing. Reinforcing consequences increase the probability of a behaviour reoccurring, whereas punishing consequences decrease the probability of a behaviour reoccurring (Skinner, 1969). Reinforcement and punishment contingencies can also be positive, meaning behaviour is affected by its presence, or negative, meaning behaviour is affected by its absence (Skinner, 1969).

Sleep is not behaviour itself (Blampied, 2013; Blampied & France, 1993). Rather, sleep is biological state that acts as a reinforcing consequence for the behaviours that occur during the phase of “falling asleep”. It is “falling asleep”

that is the behaviour under the control of discriminative stimuli that are present in the environment at the time of reinforcement (Blampied, 2013). Furthermore, sleep is a biological necessity that individuals are motivated to enter, and deprivation of sleep can increase the value of sleep, evoking the “falling asleep” behaviours (Blampied, 2013; Jin et al., 2013). Therefore, appropriate stimuli is likely the key to good sleep, and inappropriate stimuli the key to sleep problems (Blampied, 2013).

The concept of ‘behaviour chains’ is also important for understanding sleep problems (Blampied, 2013; Blampied & France, 1993). In a behaviour chain, a series of behaviours are linked together by stimuli that serve as antecedents and consequences for the preceding and proceeding behaviours (Skinner, 1969). A full bed preparation sequence is a lengthy chain, with each link under stimulus control. Choice, distraction and disruption provide possible alternative reinforcement options at each link of the chain, with the potential to disrupt the progress towards falling asleep (Blampied, 2013; Blampied & France, 1993). When stimuli that are associated with these alternative options are prominent, and their reinforcement immediate and desirable, the likelihood of interrupting the bed preparation chain is greatest (Blampied, 2013). Consistency and routine is therefore important for establishing the behaviour of “falling asleep”. The more consistent the antecedent stimuli are, the more the “falling asleep” behaviour will be associated with the stimuli, and the more embedded it will become in the individual’s behaviour repertoire (Blampied, 2013). Children are dependent on adults to provide them with the appropriate bedtime cues and reliable responses to behaviours to help them manage distractions and disruptions, and create consistent natural environments and routines in which

they can self settle every night (Blampied, 2013; Brown et al., 2014; Jan et al., 2008; Singh & Zimmerman, 2015).

According to Blampied and France (1993), there are two major requirements for the development of a positive sleep environment. First is the need for appropriate discriminative stimuli that signal and are embedded within the bed preparation behaviour chain (Blampied & France 1993). Common discriminative stimuli that encourage the likelihood of sleep in a positive sleep environment include a quiet, dark and cool room with comfortable bedding (Blampied, 2013; Jin et al., 2013). Studies have found that common rituals that parents often believe that they need to perform to get their child to sleep include co-sleeping with them, placing their child in bed already asleep following having fed, rocked, sung to, patted, walked, or driven them to sleep, or letting them fall asleep anywhere (Blampied, 2013; Blampied & France, 1993; Jan et al., 2008). As the child ages or grows, these rituals may become less desirable, and the parent may wish to change the routines. However, once these rituals have developed, they can be very hard to change as slight changes to the discriminative stimuli can disrupt sleep onset (Blampied, 2013). Furthermore, challenges may arise when the same discriminative stimuli are required every time a child needs to reinitiate sleep, including following spontaneous night wakings (Blampied & France, 1993). If these stimuli are not present when the child wakes, it can lead to the child becoming stressed and aroused, and not being able to self soothe (Blampied & France, 1993). With co-sleeping, parental presence during sleep onset can become a discriminative stimulus for sleep, and is therefore required for a child to return to sleep if they wake during the night (France & Henderson, 1996). Receiving parental attention becomes positive reinforcement for this

unsettled behaviour, strengthening the likelihood of the behaviour reoccurring. The parents are negatively reinforced themselves, as their attention alleviates the child's distress. A coercive trap is established in which both the child's behaviours and parents attention are likely to be strengthened, increased, and required to retain sleep onset (Blampied, 2013).

Secondly, Blampied and France (1993) state that contingencies of reinforcement need to strengthen and maintain sleep compatible behaviours within the behaviour chain. Disruptive nighttime behaviours not conducive to sleep, such as making demands to parents after being put to bed, and leaving the bed to seek out parent attention, may become under operant control (Didden et al., 2002). For example, they may increase if the child learns that through engaging in such behaviours they may avoid or postpone getting put to bed, and/or receive preferred items or activities as a result.

Due to deficits in social functioning, problem sleep behaviours in individuals with ASD are also likely to be maintained by variables that are not socially mediated (Campbell, 2003). These automatic reinforcers are produced by the child via their own behaviours, such as the self-stimulatory behaviours of talking to oneself, repetitive manipulation of objects, and body rocking (Didden et al., 2002; Jin et al., 2013), which normally occur when the child is alone (Hanley, Iwata, & McCord, 2003).

Changing these patterns requires a treatment informed by detailed assessment of the antecedents and consequences maintaining the behaviour, and support dealing with the negative response bursts, distress and disruption that frequently accompanies a behavioural intervention.

Common Interventions for Sleep Disorders

There are a number of approaches available for treating sleep disorders in children. The choice of which treatment to use is often, but not always, determined by the presentation of the disorder and its underlying cause (Richdale & Wiggs, 2005). Treatment strategies include pharmacological and various behavioural interventions (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006).

Pharmacological interventions. Pharmacological interventions are the most commonly prescribed interventions for children with sleep problems (Richdale & Wiggs, 2005; Vriend et al., 2011). This is due to them being a simple alternative to behaviour treatments and having an immediate positive impact on sleep (Richdale, 2013). These interventions include the use of melatonin and trimeprazine.

Melatonin. Approximately 7.2% of individuals with ASD use melatonin as a treatment for sleep difficulties (Rossignol & Frye, 2011). Melatonin is a naturally secreted neurohormone that is best known for its role in regulating the circadian rhythm (Doyen et al., 2011; Rossignol & Frye, 2011; Tordjman et al., 2013), and hence influences sleep-wake cycles. Natural levels of melatonin are commonly below average in children with ASD (Doyen et al., 2011; Rossignol & Frye, 2011; Tordjman et al., 2013), potentially due to abnormalities in melatonin-related genes (Rossignol & Frye, 2011).

Trimeprazine. Sedative medication is another widely used pharmacological intervention for sleep difficulties in children. In 2006, 16% of

New Zealand children were prescribed a sedative to help with sleep difficulties, the most common being trimeprazine tartate (trimeprazine) (Selim, France, Blampied, & Liberty, 2006). Trimeprazine is a long-acting, sedative antihistamine (www.drugs.com/mmx/trimeprazine-tartrate.html) and is dispensed in doses of 7.5mg or 30mg per 5ml (Vallergan and Vallergan forte respectively; France, Blampied & Wilkinson, 1999; Selim et al., 2006; www.medsafe.govt.nz/consumers/cmi/v/vallergratabliq.pdf).

Although pharmacological interventions can be helpful for some individuals, especially when used in conjunction with other methods, pharmacological interventions do have their drawbacks. They generally lack empirical support, and have few long-term benefits (Durand & Christodulu, 2004; Richdale & Wiggs, 2005). Undesirable side effects are often observed, such as excessive daytime sleepiness and paradoxical responses (Durand & Christodulu, 2004; Richdale & Wiggs, 2005), and there is a risk of reliance upon the medications to sleep and withdrawal symptoms when removing them (Durand & Christodulu, 2004). In addition, continual use of medication can be expensive, putting extra stress on to these families.

Behavioural interventions. Behavioural interventions are viewed as an effective alternative to pharmacological interventions (Durand & Christodulu, 2004; Weiskop, Matthews & Richdale, 2001), as they are generally more socially acceptable, and have the potential to result in more long lasting success with fewer harmful side effects (Mindell et al., 2006; Richdale & Wiggs, 2005; Vriend et al., 2011; Weiskop et al., 2001). In addition, behaviour management techniques have the potential to generalise to daytime issues (Mindell et al.,

2006), and increase parents' sense of competence, control and ability to cope (Vriend et al., 2011).

Behavioural interventions use the principles of learning theory to change how a person responds to particular stimulus (Owens, France & Wiggs, 1999). Pure behavioural interventions also involve a cognitive component, especially when working with parents to change their own behaviours, thoughts, and attitudes towards their child and routines at sleep times (Miano & Ferri, 2010; Owens et al., 1999; Weiskop et al., 2001). Behavioural interventions are described in greater detail in chapter 2.

Functional Behaviour Assessment

A large number of studies appear to assign treatments almost arbitrarily, based on the surface appearance of the problem (Brown & Piazza, 1999; Hanley et al., 2003), rather than understanding the complex combination of personally relevant antecedents and consequences that are maintaining the behaviour (Hanley, 2016). This approach means that the problem behaviour is merely 'modified, medicated, or mollified' (Hanley, 2016). Treatments that focus on changing the functional effect of the problem behaviours for that individual are likely to be more effective (Horner et al., 2002; Hanley, 2016; Campbell, 2003). An evidence-based approach that has been emerging in clinical practice and intervention literature for the treatment of challenging behaviours in children with ASD is based on Functional Behaviour Assessment (FBA; Hanley, 2016; Hanley et al., 2014; Brown et al., 2013; Didden & Sigafos, 2001; Kodak & Piazza, 2008).

Definition and process of Functional Behaviour Assessment. FBA is based on the logic of operant behaviour theory, and is a general process that aims to ascertain the discriminative stimuli and reinforcement contingencies that cause and maintain problem behaviours for an individual (Beavers, Iwata, & Lerman, 2013; Blampied, 2013; Brown et al., 2013; Hanley et al., 2014; Hanley, 2016; Horner et al., 2002; Kodak & Piazza, 2008). In FBA, outcomes from a comprehensive assessment are directly used to develop a hypothesis regarding the function of the behaviour, and tightly inform a customised treatment plan (Brown et al., 2013; Blampied, 2013; Brown & Piazza, 1999; Didden et al., 2002; Horner et al., 2002; Jin et al., 2013). Individualised treatment plans that are based on FBA are superior to generic treatments as it guarantees that the plan is based on the specific variables that are maintaining the behaviour for each individual (Minde, 1999; Spruyt & Curfs, 2015; Stores & Wiggs, 1998). A comprehensive assessment is conducted using a combination of indirect and descriptive measures (Blampied, 2013). Objective information about the antecedents and consequences maintaining the problem behaviours are obtained through interviews, ratings, checklists, self-report measures and questionnaires (Blampied, 2013). Direct measures typically include observations of the problem behaviour in the setting that it naturally occurs. Information from these sources is triangulated to build hypotheses about the factors maintaining the problem behaviours. Evidence based interventions that manipulate both the antecedents and consequences of the behaviour are specifically chosen for the individual, based on the information gathered (Blampied, 2013; Horner et al., 2002).

As a process, FBA recognises that problem behaviours are not homogenous in their etiology, and therefore it does not place an emphasis on norms, diagnostic labels, or categorising problem behaviours (Horner et al., 2002; Blampied, 2013; Brown & Piazza, 1999; Kodak & Piazza, 2008). In addition, FBA is focused on recent events relevant to the behaviour, not the past (Blampied, 2013).

Functional Behaviour Assessments for sleep problems. The process of conducting a comprehensive FBA for sleep difficulties in children with ASD involves gathering information from multiple data sources, using a variety of measures, to inform the nature of the sleep problem and the reinforcement contingencies maintaining it (Hanley et al., 2014; Blampied, 2013; Horner et al., 2002; Hanley, 2016). Common indirect methods of data gathering for sleep problems include interviews with the child's caregivers and the administration of questionnaires (Blampied, 2013). Interviews serve multiple purposes, including to gain objective information about the sleep behaviour and its antecedents and consequences, as well as the setting in which it occurs, and the frequency, intensity and duration of this behaviour (Blampied, 2013). In addition, it serves to establish information about the parents' thoughts, feelings and emotions surrounding the behaviour, their concerns, preferences, and past attempts to ameliorate the behaviour, and the child's developmental history (Blampied, 2013). It is also an opportunity to establish rapport between the family and specialist, and to ascertain the families' individual goals for treatment outcomes (Blampied, 2013). Parent-report questionnaires are included in conjunction with interviews, as a convenient and cost-effective supplement to

help categorise the type, frequency, duration and intensity of the sleep problem (Blampied, 2013). Commonly used questionnaires include the Sleep Assessment and Treatment Tool (SATT; Hanley, 2005), Child Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000), and the Questions About Behavioural Function (QABF; Matson & Vollmer, 1995) questionnaire.

Direct methods involve gathering direct observation data for the problem behaviour in the environment in which it occurs. In assessing sleep problems, this is usually achieved through parent-reported sleep diaries and all night video recordings (Blampied, 2013; Hanley et al., 2014). Sleep diaries are parent-recorded descriptions of sleep problems as they occur, and include parent's responses to their child's behaviours (Blampied, 2013). They can be used to measure the frequency, duration and setting of daytime sleeps, as well as valuable information regarding the child's sleep onset behaviours, such as the setting and time which they were put to bed, the frequency, nature and parental response to any curtain calls, and the child's sleep onset latency; the child's nighttime awakenings, including the time, duration and frequency of wakings, the child's behaviours while awake and the parents responses to these behaviours; and the time the child woke for the day. Night video recordings are used to corroborate parent's account of their child's behaviours, and capture child actions that parents are unaware of or unable to see (Jan et al., 2008; Richdale & Schreck, 2009). Direct methods help to triangulate data collected through indirect methods, avoid any parent bias, and quantify treatment effects (Knight & Johnson, 2014; Spruyt & Curfs, 2014).

A formulation based on the outcome of FBA, integrates information from all assessments to form a coherent understanding of the client's problem, and is

used to develop an individualised treatment plan that addresses the function of the sleep problem/s (Blampied, 2013). This treatment plan is often tentative, requiring adjustments during intervention. Repeated measures and observations taken throughout baseline and treatment phases allow for the analysis of different aspects of treatments, and provides opportunities to adapt and tailor the treatment according to the individual's changing needs. FBA also fits with the concept of minimal sufficiency, meaning that only the resources that are actually required to meet the optimal outcome are used. Given the ability for a FBA to tailor the treatment to an individual's needs, optimal outcomes should be reached quickly without superfluous interventions and assessments. As previously mentioned, some common antecedents and consequences maintaining sleep problems in children with ASD include a lack of sleep hygiene, parental presence during sleep onset, gaining a tangible item, and self-stimulatory behaviours. A treatment plan based on the outcomes of FBA would address these problems. For example, if the function of a child co-sleeping is hypothesised to be gaining parental attention, then a treatment plan would be developed that eliminates parental attention during sleep onset and sleep reinitiating periods.

It is important that sleep research studies measure an intervention's social validity (Finn & Sladeczek, 2001; Moore, 2004; Thackery & Richdale, 2002; Weiskop et al., 2005) as social validity has been found to correlate with treatment adherence (Brown et al., 2013) and therefore better outcomes. Brief semi-structured interviews and treatment acceptability questionnaires, for example the Treatment Acceptability Rating Form (TARF-R; Reimers & Wacker, 1992), are given to the parents post-intervention to gather valuable information, such as how the families felt about the intervention process, their understanding

about the intervention, any collateral effects on them or their child, their level of satisfaction of improvements in their child's problem behaviours, and any recommendations for future improvements (Finn & Sladeczek, 2001; Hanley et al., 2014).

Family collaboration. Children do not exist outside of a family system, but rather they impact upon and are impacted on by those around them (Bronfenbrenner, 1994; Shaffer, 2002). This ecological perspective emphasises the need to work with the families to bring about change in the child's behaviours. Due to sleep problems occurring in the home at nighttime, parents/caregivers play a critical role in implementing any treatments. Therefore, treatment plans must be socially acceptable to the families if there is to be optimal change in the child's behaviours (Turner & Johnson, 2012). A major benefit of FBA is that if desired, it allows for the incorporation of parents knowledge and preferences into the assessment process, as well as the identification of families motivations and goals (Jin et al., 2013).

FBA has a strong emphasis on modifying the antecedents and consequences of a behaviour, but can also allow for the recognition of system variables within the intervention. Often, behaviours of adults within a setting need to change to have long-lasting effects on the child's environment, and in turn their behaviour (Horner et al., 2002). For example, if a FBA determines that parental attention is maintaining the child's sleep onset latency, psychoeducation may be important to ensure that a parent changes their behaviour, and does not enter the child's room once they have put them down for the night.

Involving parents in the selection of treatment helps to alleviate any ethical or cultural dilemmas about which treatment is best practice, as different cultures may have differing sleep practices or may define sleep problems differently (Turner & Johnson, 2012). Designing interventions with family input may help to create a sense of ownership over the intervention, and may ensure that interventions are achievable and progress is maintained (Moore, 2004; Turner & Johnson, 2012).

Chapter Two

Literature Review

The purpose of this literature review is to gain a deeper understanding of the relationships between sleep problems in children with ASD and challenging day time behaviours, as well as sleep problems in children with ASD and family well-being. Evidence-based behavioural sleep interventions are discussed and are divided in to antecedent based procedures which include sleep hygiene and bedtime routines, visual supports, social stories, sensory modulation and stimulus substitution, faded bedtimes with and without response cost, scheduled awakenings and chronotherapy; and consequence-based procedures which include standing extinction, graduated extinction, minimal cheek, parental presence, and multi-modal treatment. These interventions are explored in greater detail, and the current research in these fields with children with ASD summarised, with an emphasis on intervention research that includes co-sleeping amongst its behavioural targets. Research into non-traditional approaches to treating sleep problems, such as weighted blankets, massage therapy, white noise, phototherapy, bright light therapy, restricted dieting, and herbal remedies are limited, and in some cases there is no evidence for the effectiveness of these approaches (McLay & France, 2014). Therefore, the focus of this review is on evidence-based behaviour interventions. Pharmacological interventions in combination with behavioural interventions are briefly reviewed, as there is some evidence to support this combination (for example, Cortesi, Giannotti, Sebastini, Panunzi, & Valente, 2012), and it is likely that

parents will wish to consider this multimodal method. Literature pertaining to co-sleeping in children with ASD is also reviewed, and the use of FBA in sleep interventions for this population is explored. Limitations of this research are discussed, outlining the importance of continuing research in this area.

Search Process

A systematic review of the literature was conducted, which focused on the impact of sleep problems in children with ASD on behaviour and family wellbeing, evidence-based behaviour interventions for sleep problems in children with ASD, co-sleeping, and the use of FBA for sleep problems. The following databases were included within the search: PsycINFO, PsycARTICLES, Google Scholar, ScienceDirect, and Education Resources Information Centre (ERIC). In addition, the reference lists of obtained articles, meta-analyses and systematic reviews were scanned to identify any additional articles not found on the above databases that were relevant to the review. Searches were limited to children under 18 years of age, written in the English language, and in a peer-reviewed journal. It was not limited by date, but recent articles were of a high focus.

A systematic review was conducted that focused on literature related to the impact of sleep problems on daytime behaviours in children with ASD, using diagnostic terms (“autism”, “Autism Spectrum Disorder”, “ASD”), sleep terms (“sleep”, “sleep problems”, “sleep disturbance”, “sleep difficulties”), and keywords relating to daytime behaviours and outcomes (“daytime behaviours”, “behaviours”, “challenging behaviours”, “symptomatology”, “symptoms”, “outcomes”).

A search was conducted that focused on the literature related to the impact of sleep problems in children with ASD on family wellbeing, using diagnostic terms (“autism”, “Autism Spectrum Disorder”, “ASD”), sleep terms (“sleep”, “sleep problems”, “sleep disturbance”, “sleep difficulties”), and keywords relating to family functioning and outcomes (“family functioning”, “family”, “parents”, “parents wellbeing”, “parents health”, “outcomes”).

Another search focused on literature pertaining to behaviour interventions for sleep problems in children with ASD. Search terms included diagnostic terms (“autism”, “Autism Spectrum Disorder”, “ASD”) and sleep terms (“sleep”, “sleep problems”, “sleep disturbance”, “sleep difficulties”). These search terms were then coupled with keywords related to intervention (“intervention”, “treatment”) as well as categories of intervention (“behaviour intervention”, “behaviour treatment”, “pharmacological treatment”, “medicine”).

Another systematic search was conducted that focused on literature pertaining to co-sleeping in children with ASD, combining diagnostic terms (“autism”, “Autism Spectrum Disorder”, “ASD”) with co-sleeping terms (“co-sleeping”, “parental presence”, “bed sharing”). These terms were also coupled with keywords relating to intervention (“intervention”, “treatment”). Due to the scarcity of the research relating to co-sleeping in children with ASD, a search was also conducted using only co-sleeping terms (“co-sleeping”, “parental presence”, “bed sharing”) and terms related to intervention (“intervention”, “treatment”), to provide an understanding of treatments for co-sleeping in children who did not have ASD.

Finally, a systematic search was conducted that focused on literature pertaining to the use of FBA for informing sleep problems in children with ASD,

combining keywords relating to diagnostic terms (“autism”, “Autism Spectrum Disorder”, “ASD”), sleep terms (“sleep”, “sleep problems”, “sleep disturbance”, “sleep difficulties”), and FBA (“FBA”, functional assessment”, “functional behaviour assessment”).

The Impact of Sleep Problems in Children with ASD

Children with ASD are at greater risk for sleep problems, challenging behaviours, and family dysfunction than typically developing children, yet little is known about the relationships between sleep problems and behavioural functioning, or sleep problems and family functioning in these children (Adams, Matson, & Jang, 2014; Mazurek & Sohl, 2016). If correlations between these problems exist, then early identification and treatment of sleep problems has the potential for a wide spectrum of benefits for the individual and family (Adams et al., 2014) therefore examining these relationships in young children with ASD is imperative.

The impact of sleep problems on daytime behaviour. An extensive body of literature has linked sleep problems with daytime behaviour problems in typically developing children (Mazurek & Sohl, 2016; Moon, Corkum & Smith, 2011; Sadeh, 2007). For example, Pesonen et al. (2010) explored associations between sleep duration and regularity on behavioural problems in 280 typically developing 8-year-old children. Sleep was measured with an actigraph and behaviour problems rated by parents with the Child Behavior Checklist (Achenbach & Rescorla, 2000). Pesonen et al. (2010) found that in typically

developing children, shorter sleep duration was positively correlated with more attention deficits and externalising behaviours such as rule breaking, whereas greater irregularity in sleep duration between weekdays and weekends was positively correlated with internalising problems, such as anxiety.

Previous studies have also examined these relationships in children with learning difficulties and intellectual disabilities. For example, Wiggs and Stores (1996) investigated the relationship between sleep problems and challenging behavior in 486 5 to 16-year-old children with severe learning disabilities. Sleep questionnaires were used to investigate the child's sleep, and the Aberrant Behavior Checklist (Aman & Singh, 1986) used to assess daytime behaviour problems. They found that children with sleep problems had more types of challenging daytime behaviours, and these were of a greater severity than children without sleep problems (Wiggs & Stores, 1996).

The impact of sleep on the daytime behaviour of children with ASD is a new area of research (Cohen et al., 2014; Mazurek & Sohl, 2016). Studies have begun to investigate the relationship between sleep problems and the child's ASD symptomatology as well as other daytime problem behaviours. Cross-sectional studies have found correlations between sleep problems and challenging daytime behaviours for this population. For example, Sikora, Johnson, Clemons & Katz, (2012) evaluated the association between sleep problems and daytime behaviours on a large cohort of 1193 pre-school and school-aged children with ASD, aged 4-10 years. Sleep problems were measured using the CSHQ (Owens et al., 2000), behavioural functioning assessed by the CBCL (Achenbach & Rescorla, 2000), and everyday living skills by the Vineland Adaptive Behavior Scales, Survey Interviews Form, second edition (VABS-II;

Sparrow, Cicchetti, & Balla, 2005). They found that children with ASD and sleep problems had significantly higher internalising, externalising and total scores on the CBCL than children with ASD and no reported sleep problems, regardless of age (all $p < .0001$). Furthermore, children with the greatest sleep difficulties had the greatest behaviour difficulties. However, significant relationships were found between sleep and behaviour across all ages, suggesting that age is not a factor in this relationship. Children with ASD and sleep problems also scored lower on the VABS-II as a whole ($p < .0001$), indicating a lower degree of functioning. Severity of sleep problems had no significant impact on degree of functioning ($p = .0056$).

Cross-sectional studies have also found correlations between sleep problems and ASD symptom severity. For example, Tudor, Hoffman, and Sweeney (2012) administered the CSHQ (Owens et al., 2000) and Gilliam Autism Rating Scale, second edition (GARS-2; Gilliam, 2005) to 109 parents of children with ASD aged between 3 and 18 years, to determine the relationship between sleep problems and ASD symptomatology. The GARS-2 is a parent-report questionnaire that evaluates the severity of the stereotyped behaviour, social interaction and communication domains of the Diagnostic and Statistical Manual of Mental Disorders, fourth edition (DSM-IV; APA, 2000) diagnostic criteria for ASD. Tudor et al. (2012) found that overall sleep disturbance, longer sleep onset delays and less overall sleep duration positively correlated with all ASD symptom domains as well as overall ASD severity, with sleep onset delay being the strongest predictor.

Longitudinal, cross-sectional studies have also found some relationships between sleep problems, daytime problem behaviours, and ASD symptomatology over time. For example, May et al. (2015) compared sleep

disturbance and challenging behaviours at baseline and 1 year later for 46 high functioning 7-12 year olds with ASD, and 38 typically developing children. Sleep disturbance was reported by parents on the CSHQ (Owens, et al., 2000), ASD symptoms on the Social Responsiveness Scale (Constantino, 2002), externalising problems on The Conners Third edition (Conners 3; Conners, 2003), and anxiety on the Spence Children's Anxiety Scale (SCAS; Spence, 1998). May et al. (2015) found that the ASD group had more parent reported levels of sleep disturbance at both time points than the typically developing group. However, in contrast to the control group, this level of sleep disturbance decreased over the year, but remained a significant difficulty. For the ASD group, a decrease in sleep problems was associated with both an improvement in social ability and ASD symptoms. Sleep disturbance at baseline predicted anxiety 1 year later. Aggression, hyperactivity and social difficulties correlated with sleep difficulties at both time points.

A short-term longitudinal study by Anders, Iosig, Schwichtenberg, Tang, and Goodlin-Jones (2012) found that the method of assessing sleep problems impacted significantly on the observed results. For 68 children with ASD, 69 typically developing children, and 57 children with an intellectual disability aged 2-5 years, non-subjective actigraph defined sleep problems did not relate to daytime sleepiness as defined by the Epworth Sleepiness Scale (Johns, 2015) and CSHQ (Owens et al., 2000), performance on the Bayley-III pegboard task (Bayley, 2006) or CBCL (Achenbach & Rescorla, 2000) rated challenging behaviours, but subjective parent reported sleep problems using the CSHQ and CBCL sleep question did.

Cross-sectional studies are correlational, and the aforementioned studies mostly rely on parent report and additional subjective measures, therefore making them subject to inaccuracies. Case studies are also limited, but demonstrate the promising effects of sleep interventions on daytime behaviours in children with ASD. For example, one study by Malow, McGrew, Harvey, Henderson and Stone (2006) found that total behaviour problem scores, measured using the Child Behaviour Checklist (CBCL; Achenbach & Rescorla, 2000), decreased from being in the clinical to the normal range following surgery for sleep apnoea, in a 5-year old girl with ASD. In addition, they observed increases in her social communication, noise tolerance, emotional reactivity, and alertness, as well as decreases in tactile sensitivity and repetitive behaviours. Moon et al. (2011) also found treatment to have an impact on children's behaviour problems, with two out of three 8-9 year old children with ASD having CBCL total problem scores decreasing from borderline to average following treatment for sleep problems. These changes were maintained 12 weeks post intervention.

One study was found that measured the effect of a sleep intervention on the daytime behaviours of a larger sample of children with ASD. Reed et al. (2009) recorded the sleep patterns, hyperactivity levels, and repetitive behaviours of 20 2-10 year olds with ASD before and after their parents attended a 3-part workshop on the treatment of sleep issues, using the CSHQ (Owens et al., 2000), Family Inventory of Sleep Habits (FASH: Malow et al., 2009), Parental Concerns Questionnaire (ASD specific) (PCQ: Schroeder 2014), Repetitive Behaviour Scale – Revised (RBS-R; Lam & Aman, 2007), and actigraphy. They found that following the workshop, significant improvements were found in the

children's sleep disturbances. In addition, parents reported significant improvements in hyperactivity levels, as well as self-stimulating and repetitive behaviours.

Overall, these studies demonstrate that children with ASD suffer from an increase in stereotyped behaviours, social deficits, communication deficits, and increased internalising and externalising problem behaviours when sleep is limited. Considering the correlations found between these constructs, interventions that lead to positive effects on sleep behaviours may impact on these challenging behaviour correlates.

The impact of sleep problems on family functioning. Given the prevalence and persistence of sleep disturbance among children with ASD, and the potential effect that these difficulties have on the child's daytime behaviour, the impact on the child's family can be significant. All studies that were found that looked at the relationship between sleep problems in children with ASD and family functioning were of cross-sectional design and found negative correlations between problem sleep behaviour and family wellbeing. Parents of children with ASD and sleep problems are reported to have poorer sleep quality (Lopez-Wagner et al., 2008; Meltzer, 2008; Meltzer & Mindell, 2007), higher stress levels (Doo & Wing, 2006; Hoffman et al., 2008; McStay, Trembath, & Dissanayake, 2014; Meltzer & Mindell, 2007), higher risk of depression (Foody, James & Leader, 2014; Meltzer, 2011; Tilford et al., 2015), and greater levels of fatigue (Giallo, Wood, Jellett, & Porter, 2011; Meltzer & Mindell, 2007). Siblings of children with ASD and sleep problems are also more likely to have behaviour difficulties themselves when compared to siblings of autistic children without

sleep problems (Schwichtenberg et al., 2013). Again, considering these correlations, interventions that lead to positive effects on sleep behaviours may improve family functioning.

Behaviour Interventions for Sleep Disorders in Children with Autism

Spectrum Disorder

Behaviour interventions have been widely researched than non-traditional interventions for sleep problems in children with ASD (Brown et al., 2013). Behaviour interventions are based on learning principles and the behavioural model of sleep problems (Blampied & France, 1993; Meltzer & Mindell, 2014). They are often a treatment of choice for pediatric sleep problems, as many parents prefer them to pharmacological interventions (Richdale & Wiggs, 2005; Vriend et al., 2011), they can use nonverbal means to modify behaviours, and can be tailored to individual needs and circumstances (Kodak & Piazza, 2008; Mindell et al., 2006; Owens et al., 1999; Richdale & Wiggs, 2005). Sleep problems for children with ASD have been treated with many of the same behavioural approaches as typically developing children (Mindell, 1999; Turner & Johnson, 2013; Vriend et al., 2011; Wiggs & France, 2000). However, in contrast to the plentitude of studies displaying robust evidence for the efficiency of these behavioural interventions in typically developing children, there are much fewer studies conducted with children with ASD (Schreck, 2001; Turner & Johnson, 2013; Vriend et al., 2011). In general, the studies have suggested that behavioural interventions are successful for ameliorating sleep problems in children with ASD, where the sleep problems have a behavioural basis (Turner & Johnson, 2013). A variety of behaviour intervention approaches exist, including

sleep hygiene and bedtime routines, visual supports, social stories, sensory modulation and stimulus substitution, faded bedtimes with and without response cost, scheduled awakenings and chronotherapy; and consequence-based procedures which include standing extinction, graduated extinction, minimal check, parental presence, and multi-modal treatments. Recent reviews have measured treatment efficacy using the criteria developed by Chambless and Hollon (1998). These criteria define three types of treatment efficacy: well-established, probably efficacious, and possibly efficacious (Chambless & Hollon, 1998). Different behaviour interventions, literature pertaining to its use on children with ASD, and intervention efficacies are discussed.

Antecedent- based procedures. Most treatments for sleep problems usually start with the introduction of antecedent-based procedures and the modification of sleep hygiene practices. Antecedent-based procedures are interventions based on operant behaviour theory, and use discrimination training techniques that require a stimulus to be present before the behaviour even occurs. This antecedent stimulus serves to prime the understanding that any behaviour that occurs in its presence will be reinforced. Antecedent-based procedures are also used when chaining behaviours, with each setting the occasion for the next phase of the routine. Antecedent-based procedures include establishing good sleep hygiene and a bedtime routine, visual supports, stimulus substitution, bedtime fading, sleep restriction, scheduled awakenings, and chronotherapy. Antecedent-based procedures have been used effectively with young children with ASD to ameliorate delayed sleep onset, night wakings, and

night terrors (Turner & Johnson, 2012). Sleep interventions typically begin with the establishment of positive sleep hygiene practices.

Sleep hygiene and bedtime routines. Establishing good sleep hygiene is one of the simplest components of a sleep intervention (Kodak & Piazza, 2008), and is often the first line of treatment for sleep disturbances (Jan et al., 2008). Sleep hygiene is defined as “a set of sleep-related behaviours that exposed persons to activities and cues that prepare them for and promote appropriately timed and effective sleep” (Jan et al., 2008, p 1344). Good sleep hygiene is an important contributor to sleep quality across the lifespan (Brown et al., 2014; Mindell et al., 2009; Spruyt & Curfs, 2015). Appropriate behaviours and cues for sleep onset need to be consistent, and include environmental cues (e.g. dark bedrooms a quiet environment, appropriate bed position), scheduling (e.g. regular sleep and wake times), physiologic cues (e.g. avoiding overstimulating activities before bed, adjusting the timing of meals), and positive bedtime routines (Gradisar & Short, 2013; Jan et al., 2008; Owens et al., 1999; Schreck, 2001; Vriend et al., 2011). Positive bedtime routines are a major component of sleep hygiene. They are a series of relaxing activities that the child enjoys and that are conducive to sleep (Christodulu & Durand, 2004; Kodak & Piazza, 2008; Mindell et al., 2006; Shreck, 2001), and include steps such as taking a bath, changing into pyjamas, brushing teeth, reading a book, and turning the light out (Christodulu & Durand, 2004; Schreck, 2001). Sleep hygiene can also include the removal of incompatible sleep behaviours that are maintained by reinforcement, such as television or tablet watching at night, or ensuring a child falls asleep independently (Cortesi et al., 2010; Deliens et al., 2015; Gradisar & Short, 2013).

The aim of sleep hygiene interventions are to change behaviours through increasing appropriate behaviour, rather than decreasing inappropriate behaviour, and to control affective and physiological arousal (Mindell et al., 2006). It is likely that sleep hygiene works through complex processes that combine the synchronisation of internal circadian rhythms to the external 24 hour day/night cycle, generating an association between certain activities, conditions and sleep, and creating a more calming pre-bed environment (Jan et al., 2008).

Sleep practices are significantly related to sleep hygiene in typically developing children. For example, Mindel, Meltzer, Carskadon, and Chervin (2009) ran a cross-sectional study where 1473 parents/caregivers of 0-10 year old children were asked about their child's sleep hygiene and sleep practices. They found that poor sleep hygiene was associated with poorer sleep across all ages (Mindel et al., 2009). In particular, bedtime after 9pm was associated with longer sleep onset latencies and shorter total sleep duration and parental presence was associated with more night wakings. In addition, a shorter total sleep duration was associated with an inconsistent bedtime routine, a television being in the bedroom, and regular coffee consumption (Mindel et al., 2009).

Research on establishing positive sleep hygiene practices in other developmental disabilities is scarce (Jan et al., 2008). Jan et al. (2008) wrote a comprehensive review of sleep hygiene for children with neurodevelopmental disorders that was based on their clinical experiences rather than evidence-based trials. They concluded that sleep hygiene needs to be incorporated into treatment for sleep problems in this population as a first line of treatment (Jan et al., 2008). They also recognised that sleep hygiene practices need to be modified

and adapted to meet the needs of these individuals, and that these practices are often more challenging to implement than with typically developing children (Jan et al., 2008).

Only a few studies have evaluated the effect of modifying sleep hygiene practices alone, on the sleep of children with ASD. The findings are inconsistent, with few studies finding positive effects after introducing positive sleep hygiene practices alone. In one randomised control trial (RCT) (Piazza, Fisher, & Sherer, 1997), the effects of sleep hygiene alone were compared to a faded bedtime with response cost procedure for children with developmental disabilities in an inpatient unit. Two children with ASD, aged 5 and 6 years, were included in the sleep hygiene group. These children were given a consistent bedtime routine, with designated bed times and wake times. For one child, their challenges of getting to sleep and night waking, improved slightly with just sleep hygiene practices, yet the other child, who had problems with early waking, showed no improvements.

Weiskop et al. (2005) used sleep hygiene as the first component of their intervention for five autistic children with sleep problems. Following a functional assessment, parents were educated about learning theory, and implemented bedtime routines that were tailored to their children's needs. Two weeks later, these families implemented a standard extinction intervention. For the families with autistic children, positive improvements were found in their child's settling, night waking and co-sleeping difficulties, but not until the extinction component was implemented.

In another study, Christodulu and Durand (2004) attempted to investigate the effects of establishing a bedtime routine with a 3-year-old boy

with ASD who had significant bedtime resistance and night waking difficulties, before implementing a sleep restriction procedure. However, the parents had such difficulty applying a bedtime routine due to the child's disruptive behaviours, that the sleep restriction intervention was added immediately. His behaviours improved, but the separate effects of each component could not be determined.

Adkins et al. (2012) conducted a RCT to investigate whether an educational pamphlet that included information on sleep hygiene would be sufficient to improve sleep onset latency difficulties. Parents of 36 2- to 10-year old children with ASD were randomly assigned to receive a pamphlet or not. There were no differences in sleep onset latency between the two groups following intervention, leading the authors to conclude that sleep hygiene information transmitted via a pamphlet alone is not sufficient to modify the sleep behaviour of children with ASD.

Sleep hygiene alone does not appear to be able to completely eliminate sleep behaviour problems. However, sleep hygiene needs to be addressed, or other sleep interventions are less likely to be successful (Johnson, Giannotti, & Cortesi, 2009; Vriend et al., 2011). It is considered an essential, but not sufficient, component of sleep therapy for infants and young children and is routinely included as a component of more intensive behavioural interventions (Jan et al., 2008; Mindell, Telofski, Wiegand, & Kurtz, 2009; Singh & Zimmerman, 2015; Vriend et al., 2011).

Another type of antecedent strategy that may assist in the treatment of sleep problems is the use of visual supports.

Visual Supports. Children with ASD can find it difficult to interpret verbal and nonverbal cues, and modify their behaviour accordingly (Bozkurt & Vuran, 2014; Moore, 2004; Thiemann & Goldstein, 2001). Treatment strategies that take advantage of the child's relative visual strengths and incorporate visual supports in to the intervention can be effective in changing the behaviours of children with ASD (Gray, 2010; Thiemann & Goldstein, 2001).

Social stories. Social stories are one example of a visual support used for changing behaviours in children with ASD (Bozkurt & Vuran, 2014; Moore, 2004; Styles, 2011; Test, Richter, Knight, & Spooner, 2011; Thiemann & Goldstein, 2001). Gray developed social stories in 1991 to help children with ASD learn the appropriate ways to interact in a social environment (Gray, 2010; Moore, 2004). They are visual prompts in a story format that demonstrate the appropriate behaviour responses to a particular situation (Richdale & Wiggs, 2005). Social stories are brief (between 20 and 150 words) and typically describe the settings, actions or events expected in a situation, as well as the thoughts and feelings of other people involved (Bozkurt & Vuran, 2014; Moore, 2004; Test et al., 2011; Thiemann & Goldstein, 2001). The story is written from the child's perspective, using first person language (Bozkurt & Vuran, 2014; Moore, 2004; Test et al., 2011; Thiemann & Goldstein, 2001), and should be tailored to take into account products from a detailed functional assessment, as well as the child's strengths and abilities (Moore, 2004). As many of the children receiving social story interventions can not read or have poor comprehension, social stories need to contain very simple sentences, be written in a manner that outlines what the child should do rather than what they should not do, and should contain visually stimulating and engaging pictures (Moore, 2004).

Social stories are a popular and widely used intervention strategy to help aid the comprehension of situations for children with ASD (Bozkurt & Vuran, 2014; Styles, 2011; Test et al., 2011). Reynhout and Carter (2009) conducted a survey with teachers who work with children with ASD, and found that all of them had used social stories and found them to be socially acceptable, with 93% perceiving it to be an effective support strategy. Bozkurt and Vuran (2014) put the popularity of social stories down to the fact that they are an individualisable and visually interesting mode of instruction, that can be used repeatedly and consumed at the child's own pace, are cost and time effective, easy to write and apply to varying situations, and teachers perceive them to be socially acceptable and effective. In addition, they help parents and teachers refresh what they are required to do, and encourage them to stay consistent (Moore, 2004).

Social stories have been used to increase appropriate behaviours and teach functional skills to children with ASD for a variety of challenging social situations, for example coping with grief, eating difficulties and going to hospital (Moore, 2004). Recent meta-analyses have shown that social stories are most frequently used by teachers in special education (Bozkurt & Vuran, 2014; Styles, 2011; Test et al., 2011), but can be adapted to any situation where the aim is to decrease the child's confusion and increase a desired behavioural response (Test et al, 2011).

Social stories have been used as a component of sleep interventions for typically developing children. For example. Burke, Kuhn, and Peterson (2004) effectively used a social story in combination with reinforcement to reduce bedtime resistance and night wakings in 4 2-7 year old children.

Social stories have also been used as a component of multimodal sleep interventions for children with ASD. For example, Malow et al. (2014) successfully used social stories to help children with ASD understand sleep hygiene, graduated extinction, and bedtime passes in a parent group education programme and home sleep education programme respectively. Moore (2004) also successfully used a social story as part of a multimodal intervention to help a 4 year old boy with ASD and receptive communication difficulties understand a sleep intervention that included a bedtime routine, reinforcement procedure, graduated extinction, and additional visual supports. He readily accepted all changes, and his difficulties of co-sleeping, long sleep onset latencies, frequent night wakings, and early wakings were eliminated (Moore, 2004).

Social stories are a socially valid tool that can be used to aid a child's understanding of sleep intervention. Research indicates that social stories alone do not produce robust changes and can therefore not be considered an efficacious evidence based procedure (Test et al., 2011). However they do work well as a complementary tool to more comprehensive behaviour interventions (Bozkurt & Vuran, 2014; Moore, 2004; Styles, 2011; Test et al., 2011).

Other visual supports. No literature was found that used other visual supports for sleep interventions in typically developing children. However, variants of visual supports have been used in literature with children with ASD and sleep problems. For example, Moore (2004) incorporated door hangers into a sleep intervention to help a 4-year-old boy with ASD who co-slept in his mother's bed to create distinct physical boundaries and differences between his and his mothers bedroom. Weiskop, Matthews, and Richdale (2001) and Weiskop, Richdale and Matthews (2005) both used colourful pictorial

representations of a bedtime routine on a chart to aid the understanding of changes implemented through intervention in 1-9 year old children with ASD and language difficulties, as well as settling, night waking and co-sleeping difficulties. Both studies incorporated this into an intervention that also included reinforcement procedures, modeling, standard extinction and partner support strategies. Sleep difficulties were significantly reduced and maintained at follow-up. Reed et al. (2009) also included unspecified visual supports to reinforce bedtime routines in to their parent education workshops for 22 families with 3-10 year old children with ASD and sleep problems.

One readily available tool is the 'Groclock' (see for example <http://www.thesleepstore.co.nz/shop/toolbox/trainer-clocks/general/grobaggro-clock-sleep-trainer>). This is a clock on which the face changes from a star to a sun to indicate when it is time to get up in the morning. Wake times can also be easily indicated by changing other visual aids, for example the parent may change a moon picture to a sun picture on their own or child's door at a predetermined time to let the child know it is alright to get out of bed. No research was found that investigated the use of Groclocks as a component for a sleep intervention in typically developing children or children with ASD, therefore research is required to determine its efficacy.

Sensory modulation and stimulus substitution. In addition to the core deficits required for an ASD diagnosis, many individuals also demonstrate atypical physiological and behavioural responses to sensory inputs (Ashburner, Bennett, Rodger, & Ziviani, 2013; Reynolds, Lane, & Thacker, 2011; Tomchek, Huebner & Dunn, 2014), including sensory over-responsivity (for example,

extreme reactions to taste), sensory under-responsivity (for example, apparent indifference to pain), and sensory seeking behaviours (for example feeling textures, seeking movement) (APA, 2013; Ashburner et al., 2013; Reynolds et al., 2011). Tomchek and Dunn (2007) found that in 70.5% of children with ASD, a need for sensory stimulation interfered in their daily routines, compared to 2.2% of typically developing children.

Shochat, Tzischinsky, and Engel-Yeger (2009) found that sensory modulation difficulties were a significant contributing factor for sleep problems in typically developing children, and it has been suggested that entering behavioural quietude may be a more effortful process for these children due to difficulties disengaging from a sensory environment (Milner, Cuthbert, Kertesz, & Cote, 2009; Reynolds et al., 2011). It has been hypothesised that sensory modulation difficulties may predict many sleep problems in children with ASD (Reynolds et al., 2011).

An intervention that involves decreasing or eliminating these behaviours requires a comprehensive understanding of the motivations for the behaviours, and the specific sensory qualities that are being automatically reinforced (Joosten, Bundy, & Einfeld, 2009; Patel, Carr, Kim, Robles, & Eastridge, 2000). In children who co-sleep, it is possible that the parent's presence and actions are providing them with a sensory reinforcement that aids the child to enter behavioural quietude. An intervention aimed at eliminating co-sleeping may therefore require finding an appropriate non-socially mediated stimulus substitution that replaces the parents, but maintains the same consequence. No studies were found that used stimulus substitution to aid in the elimination of co-sleeping in children with or without ASD.

Faded bedtime with/without response cost, sleep restriction. A faded bedtime procedure synchronises a child's bedtime with their sleep onset time, and then brings this forward to a more socially acceptable hour (Mindell et al., 2006). Baseline measures determine when a child will naturally fall asleep within 15 minutes of being placed in bed and a new bedtime is then set close to this time of probable rapid sleep onset. Once the child reliably falls asleep within a few minutes of being placed in bed, the bedtime is systematically brought forward until the child is falling asleep at a more desired bed time (Kodak & Piazza, 2008; Richdale & Wiggs, 2005; Turner & Johnson, 2012; Vriend et al., 2011). The child is woken at a set time each morning, and is prohibited from having day naps (Vriend et al., 2011). These interventions and their derivatives are designed to ameliorate multiple sleep related problems, such as sleep onset latency, night waking and early waking (Piazza et al., 1997).

No studies were found that used a faded bedtime procedure without response cost to treat sleep problems in typically developing children. However, it has been successful to treat insomnia in a 6-year-old girl with ADHD and a 4-year-old girl with profound mental retardation (Piazza & Fisher, 1991a). Little research has been conducted with these populations, with more focus investigating its efficacy in children with ASD.

Seven studies were found that included a faded bedtime component in the interventions for children with ASD (Christodulu & Durand, 2004; DeLeon, Fisher, & Marhefka, 2004; Durand & Christodulu, 2004; Johnson et al., 2013; Moon et al., 2011; Piazza et al., 1997; Popadopoulos et al., 2015). All of these studies aimed to improve problems of initiating and/or maintaining sleep.

Bedtime resistance problems were also addressed in three of the studies (Christodulu & Durand, 2004; Durand & Christodulu, 2004; Johnson et al., 2013).

Only one study investigated a pure faded bedtime procedure (DeLeon et al., 2004). This study aimed to decrease self-injurious behaviours associated with night waking in a 4-year-old inpatient with autism and developmental delays. The intervention stabilised his sleep patterns, decreasing the number of night wakings, and in turn decreased self-injurious behaviours.

A faded bedtime with response cost (FBRC) intervention involves a faded bedtime procedure, with the addition of increasing sleep pressure by removing the child from bed (response cost) if they do not fall asleep within the 15-minute time frame (Christodulu & Durand, 2004; Piazza et al., 1997; Vriend et al., 2011). Ashbaugh and Peck (1998) investigated the use of a FBRC procedure in a typically developing 2-year-old girl with irregular sleep patterns and co-sleeping difficulties. Using this procedure, co-sleeping was eliminated, sleep became more regular, and these effects were maintained long term.

A couple of studies were found that treated sleep problems in children with developmental disabilities with a FBRC procedure (Piazza & Fisher, 1991b; Piazza, Fisher, & Sherer, 1997). In total, 18 children between 3 and 19 years of age, in inpatient units for severe behaviour problems, with developmental disabilities that included Cerebral Palsy, Downs Syndrome, Prader-Willi Syndrome, Pervasive Developmental Disorder, Seizure Disorder, and ASD, had sleep problems treated with a FBRC procedure. Overall, most children had less disturbed sleep following treatment, with the majority showing improvement in total sleep duration, decreased daytime sleeps, decreased night wakings, and later morning wakings. In addition, Piazza et al. (1997) found FBRC to be more

effective than just implementing a scheduled bedtime. Again, although literature using the FBRC procedure in typically developing children and children with developmental disabilities other than ASD is scarce and dated, it is promising,

Two studies were found that examined the effectiveness of faded bedtime with response cost interventions in children with ASD (Moon et al., 2011; Piazza et al., 1997). Five children with ASD were included in the aforementioned study by Piazza et al. (1997), which had promising results. In the other study (Moon et al., 2011), a manualised handbook was given to parents of three 8 and 9 year old children with ASD, which included education about FBRC procedures. Moon et al. (2011) found that FBRC reduced sleep onset latency, and these improvements were maintained 3 months later.

Sleep restriction resembles FBRC, but is focused on sleep duration rather than bedtime. It involves limiting the time that the child spends in bed to 90% of their baseline total sleep time. If there is a decrease in sleep disturbances, the bedtime is gradually faded earlier until the desired bedtime is reached. If the child remains awake in bed, they are removed from their bed, and engaged in quiet activities until they appear tired.

No studies were found that investigated sleep restriction in typically developing children, but Spielman, Saskin, & Thorpy (1987) did find that sleep restriction improved sleep onset latency, sleep efficiency, and total sleep time in typically developing adults.

Durand and Christodulu (2003) and Christodulu and Durand (2004) examined the effect of sleep restriction on co-sleeping, bedtime resistance and night waking. Two children with ASD were amongst a sample of six children with developmental disabilities. No FBA was conducted and the results were mixed.

They found that the intervention eliminated co-sleeping, and decreased bedtime resistance and the frequency and duration of night wakings. However, they also showed that the participants slept less overall and one child did have an increase in the occurrence of sleepwalking and night terrors, which may have resulted from an alteration in his NREM sleep. Albeit, the parents were more satisfied with their child's sleep overall.

Overall, results of these studies have demonstrated the beneficial effects of faded bedtimes with or without response cost, and sleep restriction for sleep problems in children with ASD (Vriend et al., 2011). However, despite these positive results, these studies lacked the methodological rigor and replication required to be labeled probably efficacious (Brown et al., 2013; Vriend et al., 2011). In addition, several studies did not separate out the effects of these procedures from other components of their interventions, making it difficult to attribute results to individual factors. Furthermore, two studies (Piazza et al., 1997; DeLeon et al., 2004) used trained interventionists in inpatient clinics to implement the interventions, muddying the social validity of the intervention for being implemented in the home setting, by parents or caregivers. More research is therefore needed in to the effectiveness of these approaches to improve confidence in applying the findings.

Scheduled awakenings and chronotherapy. Additional antecedent-based procedures include scheduled awakenings and chronotherapy. When using a scheduled awakening procedure, parents preemptively wake their child approximately 15 minutes prior to the time when the child usually spontaneously awakes. When the child wakes, the parent responds as if it was a

spontaneous waking, and the child returns to sleep. The amount of time between scheduled awakenings is gradually increased, with the aim of achieving no wakings during the night and an increased duration of consolidated sleep (Durand, 2002; Mindell et al., 2006; Owens et al., 1999; Turner & Johnson, 2012). Chronotherapy involves progressively delaying the bed and wake times by a set period each day until a desired sleep time is reached (Kodak & Piazza, 2008; Owens et al., 1999; Vriend et al., 2011). During this time, other circadian cues, for example activities and meal times, are shifted too to retain a consistent and regular schedule during waking hours (Owens et al., 1999; Vriend et al., 2011). The sleep-wake cycle is systematically delayed until an appropriate sleep-wake time is established, having progressed through a 24-hour clock (Vriend et al., 2011). Scheduled awakening is limited to treating children without settling problems, and for whom parental attention is not a maintaining factor (Durand, 2002; Owens et al., 1999), and as such has been used to treat children with night terrors rather than other sleep problems (Vriend et al., 2011). Chronotherapy has typically been used to treat delayed sleep-phase syndrome, where an individual's sleep-wake phase is delayed as a result of their internal body clock being out of sync with external cues (Didden & Sigafos, 2001; Richdale, 1999). As these are not common problems in children with ASD, this literature will not be discussed in greater depth.

The literature for antecedent-based interventions for sleep problems in children with ASD is promising, with improvements seen in co-sleeping, sleep onset latency, night wakings, early wakings and bedtime resistance. Given the scarcity of research however, these techniques cannot yet be classified effective.

Antecedent interventions are often perceived to be essential, but not sufficient components of sleep interventions, and are frequently used in conjunction with consequence-based interventions.

Consequence-based procedures. Consequence-based procedures focus on manipulating the factors that occur directly after a problematic behaviour occurs, in an effort to decrease the likelihood of that behaviour reoccurring (Wiggs & France, 2000). Consequence-based procedures include standard extinction, graduated extinction, minimal check, and parental presence.

Standard extinction. Standard extinction procedures to address sleep behaviour problems require an adult to consistently withhold reinforcements that have been a reliable consequence for undesired behaviours which occur when the child is meant to be sleeping (Didden et al., 2002; Owens et al., 1999; Turner & Johnson, 2012; Vriend et al., 2011). For example, when it has been established that a child calling out when in bed at night is being maintained by receiving adult attention, standard extinction would consist of the adult ignoring the child's behaviour and not attending to the child until the morning, unless the parents deem it to be absolutely necessary (Owens et al., 1999; Turner & Johnson, 2012; Vriend et al., 2011). As a consequence of the behaviour no longer resulting in the desired reinforcement, the problem behaviour is decreased or eliminated (Vriend et al., 2011).

Standard extinction is a method commonly used for treating settling and night waking problems in typically developing children, and has well-established efficacy for this population (Mindell, 1999; Weiskop et al., 2005). Single case and

group studies now exist that show standard extinction has a rapid and positive effect on sleep onset latency and night waking difficulties in children with a range of disabilities. For example, Didden et al. (1998) conducted case studies on six 2-7 year old boys with sleep problems and either Prader-Willi syndrome, Fragile-X syndrome, ADHD, spastic tetraplegia, spinal muscle atrophy or spastic diplegia. These children had a range of sleep problems, including co-sleeping, settling difficulties, sleep onset latency, night wakings and bedtime resistance. A FBA was conducted to determine the factors maintaining each individual's problems. Standard extinction was used in each case, and all children had improved sleep behaviours following intervention. Thackery and Richdale (2002) used standard extinction to treat sleep behaviours in three 5-10 year old boys with intellectual difficulties. Before intervention, these children all required a parent's presence to fall asleep, two co-slept, and two had frequent night wakings. Following intervention, all children fell asleep independently, and did not co-sleep, and night wakings were reduced in one child, with effects maintained long-term. Research suggests that standard extinction is an effective tool for treating sleep problems in typically developing children and children with developmental disabilities.

Four studies were found that examined the effects of standard extinction procedures to treat sleep problems in children with ASD. Both Wolf, Risley and Mees (1964) and Didden et al. (2002) used FBA's to determine that parental attention was maintaining bedtime resistance and night wakings in their participants: a 3-year-old boy with ASD, and a 6-year-old boy that was included amongst four children with developmental disabilities respectively. Parents were instructed to extinguish all attention during the night. Both of these

children showed a rapid and significant reduction in their problem behaviours that were maintained at a 6-month follow-up.

Weiskop et al. (2001; 2005) conducted two studies that incorporated standard extinction in to behaviour interventions for children with ASD and sleep problems. A case study of a 5-year-old boy lead to a wider study with 13 1-9 year old children, six of whom had ASD. In both studies, these children had difficulties that included bedtime resistance, night wakings and co-sleeping. Following a FBA, parents were given three individualised training sessions that focused on educating them around sleep hygiene, reinforcement and standard extinction techniques. Improvements were seen in the children's co-sleeping, bedtime resistance and night wakings that were maintained at follow-up. Inconsistent results were seen for sleep onset latency and sleep duration. Tracking the children's behaviours evidenced that implementation of standard extinction lead to a quick change in the children's behaviours.

Overall, these studies show that standard extinction can rapidly and effectively decrease co-sleeping, night wakings, and bedtime resistant behaviours in children with ASD. Given that standard extinction consistently improved sleep in more than three children in methodologically sound studies, it meets the criteria for a possibly efficacious intervention for this population (Vriend et al., 2011).

Post-extinction response bursts. Despite standard extinction procedures having a rapid and marked effect on the reduction of challenging behaviours in both children with and without ASD, these procedures can create an undesirable side effect known as a Post Extinction Response Burst (PERB). As described by

Skinner (1964), when a challenging behaviour is first put under extinction, there is often a temporary increase in the intensity, duration, frequency and range of behaviours before it improves (Didden et al., 2002; France & Blampied, 2005; Kodak & Piazza, 2008; Richdale & Wiggs, 2005; Owens et al., 1999; Skinner, 1969; Turner et al., 2012; Vriend et al., 2011). For example a child who is first denied any parent attention while getting to sleep may attempt to maintain their attention through more aggressive means such as screaming and crying longer and louder (Turner et al., 2012). Following a PERB, it is typical to have a period of settled behaviour, followed by a temporary spontaneous recovery of PERB-like behaviours.

This PERB creates a potentially challenging situation, as the increase in unwanted behaviours can cause distress to both the children and the parents (France & Blampied, 2005; Kodak & Piazza, 2008; Owens et al., 1999; Turner et al., 2012; Vriend et al., 2011). Procedures may become difficult for parents to consistently adhere to when faced with a rise in difficult behaviours (France & Blampied, 2005; Owens et al., 1999; Turner et al., 2012), making them prone to abandoning the extinction procedure in order to attend to their child (Turner et al., 2012). Unfortunately, attending to a child during the PERB inadvertently reinforces the child's behaviour at this intensified level, making it more likely that the child's behaviour will reoccur at a heightened strength, making the behaviour more difficult to change (Kodak & Piazza, 2008).

A study by Price, Wake, Ukoumunne & Hiscock (2012) found that no long-term harm is caused to a child put in to an extinction procedure. Nevertheless, parents can be unwilling to use a standard extinction method due to fears of psychological trauma that they believe may result if they ignore their child's

distress (France & Blampied, 2005; Owens et al., 1999; Singh & Zimmerman, 2015; Vriend et al., 2011).

Given that children with ASD often have multiple challenging behaviours aside from sleep problems, such as aggression, self-injurious behaviours and emotional outbursts, extinction procedures may not be a viable treatment option for many families (Hanley et al., 2014; Kodak & Piazza, 2008; Lancioni et al., 1999; Liu et al., 2006; Singh & Zimmerman, 2015; Wiggs & France, 2000). A PERB may increase these displays of challenging behaviours, and therefore raise safety concerns for the child or other family members (Kodak & Piazza, 2008; Wiggs & France, 2000).

Overall, when implemented correctly, standard extinction procedures are found to be effective and rapid treatments for sleep difficulties (Didden et al., 2002; Kodak & Piazza, 2008; Turner & Johnson, 2013; Vriend et al., 2011). However, given the challenges that arise from standard extinction procedures, treatment adherence and social acceptability is sometimes low, and standard extinction is not a viable treatment option. This has led to researchers investigating alternative, less restrictive approaches to treatment (Blampied, 2013; McLay & France, 2014; Vriend et al., 2011).

Modified extinction procedures. In order to retain the effectiveness of standard extinction procedures, but improve their social acceptability and adherence levels, modified versions of extinction procedures have been developed (Lerman, Iwata & Wallace, 1999; Vriend et al., 2011). Modified extinction procedures of graduated extinction, including minimal check and parental presence are discussed.

Graduated extinction. A graduated extinction procedure is one in which challenging behaviours are gradually decreased through incremental withdrawal of the reinforcer over time (Singh & Zimmerman, 2015; Wiggs & France, 2000). For example, if parent attention is maintaining a prolonged sleep onset time, parents either systematically increase the length of time they take before responding to bed-time crying, or systematically decrease the time spent interacting with the child during settling (Kodak & Piazza, 2008; Mindell et al., 2006; Owens et al., 1999; Turner & Johnson, 2012). Lawton, France & Blampied (1991) suggest that a baseline level of the duration of the reinforcement, for example time spent responding to the child, is established and then incrementally reduced in duration by 1/7th every four nights. The goal of graduated extinction is to gently teach the child to develop “self-soothing” skills so that they can independently fall asleep without their undesirable sleep associations (for example parents attention, milk bottle) (Knight & Johnson, 2014; Mindell et al., 2006). Once established, these skills should generalise to normal night wakings, and the child should sleep through the night (Mindell et al., 2006).

Many studies have been conducted that have shown graduated extinction to be an effective intervention for sleep problems in typically developing children (for example, Adams & Rickert, 1989; Lawton et al., 1991; Moore, 2004), suggesting it is as equally efficacious as standard extinction (Kuhn & Elliott, 2003; Richdale & Wiggs, 2005). For example, a recent study by Moore (2010) found that treating a 3-year-old girl with graduated extinction in combination with sleep hygiene practices, significantly improved her presenting problems of co-sleeping, frequent night waking and long sleep onset latencies. The efficacy

and social acceptability of this approach has made it a popular intervention. In fact, in Australia, 52% of pediatricians use graduated extinction to treat night wakings in typically developing children (Heussler et al., 2013).

Research in to the use of graduated extinction in children with developmental disabilities is ongoing, with results suggesting it to be a viable and possibly preferable treatment plan for this population too. Durand, Gerner-Dott and Mapstone (1996) used graduated extinction in conjunction with establishing a bedtime routine to address co-sleeping, settling problems and night wakings in four children with developmental disabilities, aged 2-12 years, two whom had a diagnosis of ASD. A FBA was conducted and interventions individualised. Sleep problems were reduced in all children, including the elimination of co-sleeping, with improvements maintained at a 6-month follow-up assessment. Parents reported the treatment was easy to implement and adhere to (Durand et al., 1996).

Many years ago, Howlin (1984) used graduated extinction as the primary intervention to successfully reduce co-sleeping, night waking and night settling problems in a 5-year-old boy with ASD, by gradually increasing the distance which the mother slept from the child. In addition, the mother reported her own mood improvements, evidenced by no longer relying on the use of anti-depressants, as well as improvements in her marital relationship as a result of the intervention (Howlin, 1984).

Although improvements were seen in all these children following graduated extinction, more methodologically rigorous studies are required before it can be deemed a possibly efficacious treatment for this population (Vriend et al, 2011).

Minimal check. One variation of a modified extinction method is a minimal check procedure, which is an intensive, temporary method that involves the parent checking the child at regular intervals throughout an extinction procedure (France & Blampied, 2005; Owens et al., 1999; Richdale & Wiggs, 2005). Parents behaviour is altered in that their responses are not contingent on the child's behaviour, rather they wait and respond to their child's waking at successively longer durations of time, ignoring all other bids for attention (France & Blampied, 2005; Owens et al., 1999; Turner & Johnson, 2013). Intervals between checking have ranged from 5-20 minutes (Owens et al., 1999). At each check, the parent will attend to the child for a set duration, during which the parent provides minimal attention to the child, restoring their sleep position and offering verbal assurance only where needed, before leaving the room again (France & Blampied, 2005).

Minimal check has been found to successfully reduce problem sleep behaviours in typically developing children, with little risk of a PERB (France & Blampied, 2005; France, Blampied, & Henderson, 2003; Matthey & Grncec, 2012). One case study was found that used a minimal check procedure with a child with a developmental disability other than ASD. O'Reilly, Lanciono, and Sigafos (2004) reported on a case of a five-year-old girl with severe intellectual disabilities and difficulties settling. The authors hypothesised, from a functional analysis, that her behaviours were maintained by her mother's presence. A bedtime routine and a minimal check procedure where the mother provided attention for 20 seconds every 5 minutes was used. Sleep latency and curtain calls were decreased and these effects were maintained 12 months later. A

unique aspect of the study was that a reversal design was able to show these improvements were attributed to the minimal check procedure.

In 2004, Moore used a minimal check procedure in conjunction with a social story to treat a 4-year-old boy with ASD and co-sleeping, sleep onset latency and night waking difficulties. Following a FBA, it was found that the function of his sleep difficulties was to gain attention, particularly from his mother. When he called for her during the night, she was instructed to initially delay attending to him for 1 minute, and increase this delay by 1 minute each night. When attending she provided him with minimal affection and verbal reassurance. The mother reported that this intervention was easy to implement and adhere to. Results were unclear however. It appears that this treatment eliminated his co-sleeping, but nothing was reported about the impact it had on his night wakings and sleep onset latency. Furthermore, no long-term follow-up was conducted.

Parental presence. Another way to gradually withdraw parental responses that are maintaining problem behaviours is an approach called parental presence (France & Blampied, 2005; Mindell et al., 2009; Owens et al., 1999; Stores, 1996). This procedure is generally used when a functional assessment determines sleep problems are being reinforced by parental presence (Owens et al., 1999), and is therefore commonly used when the goal is to decrease co-sleeping, sleep onset latency, and night wakings. In this procedure, the parent remains in the child's room, on a separate bed or chair, until the child falls asleep (France & Blampied, 2005; Owens et al., 1999). The parent sleeps in the child's room but does not interact with the child even if they wake, unless deemed

absolutely necessary for safety reasons (France & Blampied, 2005; Owens et al, 1999). Should the parent need to engage with the child, interaction is kept to a minimum, for example a parent may restore a child's sleeping position, but not interact verbally (France & Blampied, 2005). Parental presence is thought to provide the child with reassurance and allow them to settle without any anxiety, at the same time as reinforcing attention given by the parent is removed (Sadeh, 1994). After the child is able to settle and resettle independently, the parent's presence is removed altogether (France & Blampied, 2005; Owens et al, 1999).

A variation of parental presence is a 'camping out' technique, where the parent leaves the room once the child is asleep. Again, removal of the parents' presence is systematically faded out until their presence is no longer required for the child to self-settle (Papadopoulos et al., 2015). For example, in this procedure, the parent may start sitting on the child's bed until they fall asleep. Contingent on the child's behaviour, the parent 'camps out' in the child's room, sitting in positions incrementally further away from the child's bed until the presence of the parent is fully eliminated. Should the child seek the parent's attention during the night, the parent returns them to bed and 'camps out' in the same position that they did during initial sleep onset, until the child is asleep.

Parental presence is emerging as an effective method for reducing sleep problems in typically developing children, with a decreased PERB (France & Blampied, 2005; Matthey & Crucec, 2012; Mindell, 1999). For example, in a large group study, Sadeh (1994) found parental presence to be comparably efficacious to a minimal check procedure. In a more recent example, Moore (2010) found that a parenting presence procedure lead to improvements in the sleeping behaviours of a 3-year-old girl, decreasing her night wakings, and eliminating co-

sleeping. In Australia, 21% of pediatricians use parenting presence as a tool to treat sleep initiation difficulties, and 18% use it to improve frequency of night wakings (Heussler et al., 2013).

One study was found that investigated the use of a parenting procedure in children with developmental disabilities other than ASD. Hewitt (1985) used parental presence to address settling and night waking difficulties in ten children with severe intellectual disabilities. Parents gradually moved further away from their child during sleep onset initiation and following a night waking. Seven of the children had improvements in their sleep behaviours, which were maintained at a 12 month follow-up.

Given the high levels of anxiety observed in children with ASD, parental presence could be a viable and less anxiety-provoking treatment for eliminating sleep behaviours maintained by parent attention. However, only one study was found that used parental presence for this population. Howlin (1984) conducted a single case study using parental presence to address co-sleeping, settling and night waking problems in a 6-year-old boy with ASD. Over an 8 week period, the mother gradually moved from co-sleeping on a mattress next to his bed, to returning to her own bed. Co-sleeping and settling problems were eliminated, and night wakings reduced, with gains maintained at a 6 month follow-up. In addition, the mother's wellbeing improved, and she was able to stop taking antidepressants that were initially prescribed to her because of the stress from her son's behaviour.

In a unique study conducted by France and Blampied (2005), the authors compared standard extinction with these modified extinction procedures in

typically developing children with sleep problems. They found all procedures decreased night wakings. Standard extinction procedures had the greatest PERBs but the quickest resolution of night wakings. Minimal check procedures were more variable in their results between participants, and appeared more gentle due to fewer PERBs, but actually showed more crying as the intervention progressed, and poorer resolution of awakening. Parental presence procedures, however, showed characteristic PERBS in only a minority of the studies. All parental presence studies resulted in a rapid decrease in waking and crying. They concluded that parental presence procedures were the treatment of choice due to a decreased likelihood and magnitude of a PERB, but still leading to dramatic and robust changes in night wakings. It has been suggested that parental presence procedures are preferred for children with ASD, as gradual removal of reinforcement provides the child with a chance to adjust to the new routine, minimizing distress and allowing for tailoring to the child's individual pace of learning (Kodak & Piazza, 2008; Stores & Wiggs, 1998). Research in this field with children with ASD is still in its early stages, and more research needs to be done to investigate these specific techniques with children with ASD (Vriend et al., 2011).

Multimodal behavioural treatments. Over the last 20 years, several studies have used multimodal approaches to treatment (varying combinations of extinction, graduated extinction, sleep restriction, faded bedtime, sleep hygiene, visual supports and reward systems) for children with ASD and sleep problems.

Bartlet and Beaumont (1998) included seven individuals with ASD in their research that used extinction procedures in conjunction with sleep hygiene,

graduated extinction and positive reinforcement to target settling, night waking and early waking difficulties in children with ASD aged 11-27 years. No functional assessment was conducted, and the parents of 45/57 participants reported improved nighttime behaviours, with the remaining participants staying the same or even worse.

Montgomery, Stores and Wiggs (2004) conducted the first RCT in this field with 66 2-8 year olds, 21 with ASD, who had bedtime resistance, night waking and co-sleeping difficulties. Participants were allocated to groups receiving either education around extinction and modified extinction methods via a simple booklet, or through face-to-face dissemination of the strategies provided in the booklet, or given no intervention for the first 6 weeks. No functional assessment was conducted. Both treatment groups were equally effective when compared to controls, with 2/3 of the children showing significant improvements in their sleep onset latency and night wakings that were maintained after 6 months.

Another study to incorporate multimodal techniques in to an education programme for this population was conducted by Malow, MacDonald, Fawkes, & Alder, (2016). In a single case design, the parents of eight 3-9 year old children with ASD and sleep difficulties including bedtime resistance, sleep onset latency, night wakings and co-sleeping, were given a sleep manual which covered the behavioural techniques of standard extinction, bedtime routines, bedtime pass and visual schedules, and then left to implement these tools with their children without any guidance. No functional assessment was conducted. Three quarters of the children showed improvements in their sleep behaviours. In particular, 4/6 decreased their sleep onset latency, 4/7 had improvements in their number

of night wakings, 3/6 improved their bedtime resistance, but only 2/5 improved the frequency of co-sleeping.

Reed et al. (2009) ran three 2-hour group workshops that provided psychoeducation around establishing good sleep hygiene, positive bedtime routines, and reinforcement procedures to 20 families with 3-7 year old children with ASD. No FBA was conducted. Before the workshops, these children presented with combinations of bedtime resistance, night wakings, early waking, and co-sleeping. Subjective reports suggested that the treatment significantly improved sleep onset latency, sleep duration, bedtime resistance, and sleep anxiety, but night wakings did not improve significantly, and still persisted for most children. Five out of seven children with co-sleeping issues at baseline reported fewer nights of co-sleeping following the workshop. Actigraph results showed an objective improvement in sleep onset latency, but mixed results for night wakings for children presenting with these difficulties.

Knight and Johnson (2014) also provided their participants with a multimodal package of behaviour interventions. Three children with ASD, aged 4-5 years, who had difficulties with sleep onset latency and night wakings were asked to do a one-month long intervention where they used sleep hygiene, positive bedtime routines, white noise and graduated extinction. No FBA was conducted. Subjective measures showed all children decreased their sleep onset latency and night waking frequencies, which were maintained at follow-up.

Malow et al. (2014) conducted a RCT with 80 2-10 year olds with ASD, to determine if group or individual parent education would impact on sleep onset latency difficulties. Participants were randomly allocated to either group education, where two to four parents engaged in two 2-hour sessions, or

individual education where parents were given two private 1 hour sessions. These sessions focused on psychoeducation around sleep hygiene, bedtime routine, graduated extinction, bedtime passes, and social stories. Results showed that mode of education did not affect outcomes, with decreases in sleep onset latency seen in both groups.

Two larger RCT's incorporated faded bedtimes in to their intervention (Johnson et al., 2013; Papadopoulus et al., 2015). Johnson et al. (2013) delivered a five session one on one manualised behavioural parent training programme to 33 parents of children aged 2 to 6 years with ASD and bedtime resistance, sleep onset latency, night wakings and/or early waking difficulties. Effects were compared to families who were given non-sleep related parent education. Papadopoulus et al. (2015) tailored behavioural sleep management plans to 61 5- to 13-year old children with comorbid diagnoses of ASD and ADHD, and compared these to participants who continued with usual clinical care by pediatricians. Both studies found that subjective reports of sleep problems improved following treatment for the intervention groups. Papadopoulus et al. (2015) found that these results were maintained at 3 and 6 month follow-ups.

Multimodal behavioural approaches to treatment for sleep problems have resulted in significant improvements for children with ASD. Given that a package of treatments were presented simultaneously in most studies, effects of individual treatment components were not teased apart. In addition, the majority of these studies did not conduct a FBA to determine the specific variables maintaining the children's sleep problems. This leads to uncertainty about which components of a multimodal package are required for change and which are most effective.

Pharmacological interventions. Pharmacological interventions, in particular melatonin and trimeprazine, are often used to treat sleep problems in children, as they are simple to implement and have immediate effects (Richdale, 2013). The use of melatonin is found to be efficacious for sleep difficulties in typically developing children (Doyen et al., 2011; Rossignol & Frye, 2011), but has had mixed results for children with ASD (Doyen et al., 2011; Malow et al., 2012; Paavonen et al., 2003; Rossignol & Frye, 2011; Wright et al., 2011).

More than one third of clinicians recommend the use of melatonin for treatment of sleep problems in children with developmental disabilities (Schwichtenberg & Malow, 2015). Despite its common use, relatively few studies have documented its efficacy in children with developmental disabilities. Several recent meta-analyses of studies that used melatonin as the primary intervention for sleep difficulties in individuals with ASD have found that studies consistently find a significant decrease in sleep onset latency with its use (Doyen et al., 2011; Malow et al., 2012; Rossignol & Frye, 2011; Wright et al., 2011). About half of the studies report an increase in total sleep duration, while the other half show no differences in comparison to a placebo (Schwichtenberg & Malow, 2015). Results have been mixed for its effect on night wakings, with some (for example, Paavonen et al., 2003) showing an increase in night wakings following melatonin treatment. These results suggest that melatonin is good for treating sleep onset difficulties in this population, but cannot be relied on for other sleep problems.

Other studies have found some negative outcomes associated with the use of melatonin in individuals with developmental disabilities. For example, melatonin has been found to produce an initial positive effect that waned over time, despite an escalation in dose (Rossignol & Frye, 2011; Tordjman et al.,

2013). Also, in most studies, discontinuation of melatonin led to a return to pre-treatment sleep behaviours (Doyen et al., 2011; Paavonen et al., 2003).

Research into the use of trimeprazine has found that it provides some short-term relief, but inconsistent and non-clinically significant changes in sleep difficulties have been found long-term in young children treated with trimeprazine alone (France et al., 1999; Richman, 1985).

Pharmacological and behavioural interventions combined. Several researchers have investigated the effects of combining pharmacotherapy with behavioural interventions, with the rationale being that the short-term sedative effects of the medications would soften the PERB typically exhibited at the beginning of an extinction treatment (Cortesi et al., 2012; France et al., 1991; Selim et al., 2006). The child would still learn the long-lasting fundamental lessons that a behavioural intervention teaches, but the procedure would be less distressing for the child and less stressful for the parent, therefore making it more acceptable to the family (Cortesi et al., 2012; France et al., 1991; Selim et al., 2006). In addition, parents reluctant to use medications would be able to wean their child off the medications and still have lasting improvements (Selim et al., 2006).

In 1991, France et al. conducted a double-blind placebo-controlled study investigating the effects of decreasing doses of trimeprazine in combination with planned ignoring on the sleep disturbance of typically developing children. They found that all children decreased their frequency and duration of night wakings to low levels which were maintained long term. However, in contrast to controls who were prescribed extinction or extinction and placebo treatments, children

who had the combined treatment plan had more abrupt decreases in night wakings, and decreased PERBs. In addition, all groups significantly improved infant security ratings, and decreased levels of maternal anxiety.

Selim et al. (2006) modified this procedure by adding a parental presence component, and found similar results, concluding that even though Trimeprazine alone is rarely associated with long-term improvements in sleep difficulties, it does have positive effects when used in conjunction with behavioural interventions (France et al., 1991; Selim et al, 2006).

Combining behavioural interventions with melatonin or trimeprazine appears to result in more rapid improvements in sleep behaviours that are clinically significant and more acceptable to families with and without ASD than extinction or graduated extinction procedures alone.

Summary of interventions. To summarise, the majority of studies investigating treatments for sleep problems in children have focused on behaviour interventions (Meltzer & Mindell, 2014; Turner & Johnson, 2013). Given the increased likelihood of cognitive and language difficulties, and the heterogeneity between individuals with ASD, behaviour interventions may be particularly suited to children with ASD, as they use non-verbal means to modify behaviours and can be tailored to the individual's needs and circumstances (Kodak & Piazza, 2008; Mindell et al., 2006; Richdale & Wiggs, 2005; Turner & Johnson, 2013). Robust evidence exists that demonstrates the effectiveness of a range of behaviour interventions for improving sleep in typically developing children (Meltzer & Mindell, 2014). However, research in to behavioural treatment for sleep problems in children with ASD is still in its infancy. No

interventions have met the criteria to be classified as “well-established” (Vriend et al., 2011). The individual efficacy of treatment components is not fully known, but cumulative effects are promising (Montgomery et al., 2004). Overall, these studies have a range of methodological limitations, such as small sample sizes, and variations in study designs, design quality and choice of outcome measures. This restricts the certainty and generalisability of the results. Not all studies included data gathered during baseline, intervention and follow-up phases. These phases are important methodological components of a research study, that helps analyse the impact of the treatment as a whole and of its individual components, in the short and long-term. Reporting of information regarding participant characteristics is inconsistent (Turner & Johnson, 2012; Vriend et al., 2011). Given that populations of children with ASD and sleep difficulties are heterogeneous in many aspects, including their ASD symptoms and severity, comorbid conditions, cognitive and developmental abilities, presenting problems, histories, and ecological circumstances, these aspects need to be considered when selecting interventions. For example, non-verbal children may require more visual supports than those with language (Turner & Johnson, 2012), and children with anxiety or depression co-morbidities may need to have these issues addressed first. Reports are encouraging, but there are an insufficient number of studies into all areas of behaviour interventions for children with ASD. Studies need replication and to be done with greater methodological rigor to increase confidence in implementing these interventions.

Co-sleeping

Co-sleeping is often used by parents as an effective tool for reducing sleep problems, in particular bedtime resistance, long sleep onset latencies, and parent seeking following night wakings (Richdale, 2013). However, it can become a problem when parents wish to establish more independence in their child, or it is significantly disrupting their sleep and wellbeing too.

A limitation in the research on co-sleeping is that it is usually conducted in typically developing infants who are co-sleeping intentionally (Goldberg & Keller, 2007). Most literature has discussed the prevention of co-sleeping. No studies were found that addressed the treatment of solely co-sleeping problems in typically developing children, most probably because co-sleeping is normally not an isolated sleep problem, but often a consequence of other sleep difficulties. No studies were found that addressed treatments for solely co-sleeping issues in children with developmental disorders either. Some studies with children with ASD have included co-sleeping as one target of treatment in this population, and these have been described in greater detail in this thesis already (for example Durand & Christodulu, 2004; Howlin, 1984; Malow et al., 2016; Montgomery, Stores, & Wiggs, 2004; Moore, 2004; Reed et al., 2009; Weiskop et al., 2001; Weiskop et al., 2005).

Due to the high prevalence of reactive co-sleeping in children with ASD, more research is required to develop treatment protocol for these families.

Functional Behaviour Assessment

The impact of Functional Behaviour Assessment on problem

behaviours. One way to determine the most appropriate antecedent and consequence based treatments to use is FBA. FBA for problem behaviours are well researched (Hanley, 2016). A review conducted by Beavers et al. (2013) found 435 published studies that used FBA. The literature has shown that larger reductions in problem behaviours are found when FBA is used to inform the intervention than when a generic intervention is used (Beavers et al., 2013; Campbell, 2003; Hanley, 2016; Hanley et al., 2003; Horner et al., 2002;), and the more precise the assessment, the higher the success rate (Horner et al., 2002). Despite these findings, few interventions are based on FBA, and even when FBA is done, interventions are being used that contradict the information gained (Campbell, 2003; Horner et al., 2002).

In children with ASD, FBA has been shown to inform simple and effective interventions for problem behaviours, such as tantrums and self-injury (Campbell, 2003; Hanley et al., 2014; Hansen & Wadsworth, 2015). For example, Hanley and colleagues (2014) used FBA to inform intervention for three 3-11 year old children with ASD, who all had problem behaviours that included aggressive tantrums, that occurred multiple times per day. Hanley et al. (2014) used multiple assessment measures to identify the antecedents and consequences that maintained these children's behaviours, in order to tailor interventions to each child that would decrease these behaviours. Specific functional communication responses to replace the tantrum behaviours were taught to each child, followed by denial- and delay- tolerance training. After

intervention, none of the target behaviours were observed, and skills were found to generalise to other setting and situations (Hanley et al., 2014).

A recent single case study by Hansen and Wadsworth (2015) used FBA to inform intervention for a 10-year-old boy with ASD, intellectual disability, hearing impairment and a stigmatism, who had repetitive behaviours of eye poking and clapping. A FBA determined that both behaviours were maintained through automatic reinforcement, leading to an intervention involving giving the boy access to a choice of items to hold that provided him with visual and tactile stimulation, and were incompatible with the self-stimulating behaviours (Hansen & Wadsworth, 2015). Eye poking was eliminated, and hand clapping significantly reduced, with these gains maintained at a 9-month follow-up (Hansen & Wadsworth, 2015).

Functional Behaviour Assessment, sleep, and ASD. FBA is well suited as a means to inform sleep interventions for children with ASD, as it links a comprehensive assessment to an individualised treatment plan (Brown et al., 2013; Hanley et al., 2014; Kodak & Piazza, 2008). Given the heterogeneity of symptoms and associated features of children with ASD, and the varying etiologies of their sleep disturbances, there is a heightened need to tailor interventions specifically for the individual's needs (Horner, 2002; Campbell, 2003).

FBA is only just beginning to be used as a tool to synthesise assessment and evidence-based treatments for children with ASD and sleep problems (Hanley et al., 2014; Jin et al., 2013). For example, Papadopoulos et al. (2015) used FBA to inform hypotheses about the factors maintaining sleep problems in

28 5-13 year olds with comorbid ASD and ADHD. These children had a variety of sleep problems, that included sleep onset difficulties, night wakings, and sleep anxiety (Papadopoulos et al., 2015). Information gained through consultations, sleep diaries and the CSHQ (Ownes, Spirito, & McGuinn, 2000) lead to individualised treatment plans tailored to the families needs, for example graduated extinction was used to decrease the need for parental presence, and sleep restriction was used to shorten lengthy sleep onset latencies (Papadopoulos et al., 2015). Mid to large improvements were found in the sleep problems compared to a care as usual control group, and these were maintained at a 6-month follow up (Papadopoulos et al., 2015). In addition, small to moderate improvements were found in non-target areas, including the children's psychosocial quality of life, ADHD symptom severity, and daytime behaviours (Papadopoulos et al., 2015).

Jin and colleagues (2013) described their FBA process for determining a treatment plan for three 7-9 year old participants who had developmental disabilities (two with ASD) and sleep problems. Jin et al. (2013) used sleep diaries and infrared nighttime videos to obtain data about the children's sleep onset, sleep interfering behaviours, night wakings, total sleep, parental presence and use of medication. In addition, they developed and used the Sleep Assessment and Treatment Tool (SATT), which is an open-ended parent interview that gains insight in to the types of sleep problems their child is currently experiencing, and identifies parents goals as outcome measures. One child was identified as having delayed sleep onset with sleep interfering behaviours. To decrease his sleep onset latency, a faded bedtime without response cost was used. The FBA determined that his sleep interfering

behaviours were being maintained by access to parental attention and tangible items. Based on this information, treatment included providing the child with concentrated access to his parents and the desired items for a period before sleep, and then restricting them at bedtime. The second child was identified as having delayed sleep onset, and lengthy night wakings. Sleep interfering behaviours were hypothesised to be maintained by automatic reinforcement, and therefore treatment included allowing the child to engage in this behaviour before bed, and then restricting and interrupting this behaviour after bedtime. This child fell asleep to music, but this music was turned off after during the night. Lengthy night wakings were hypothesised to be due to this child's association between music and sleep onset, with these conditions not being met during the night. Music was eliminated at bedtime and throughout the night, ensuring sleep conditions remained consistent. The third child was identified as having difficulties settling, frequent night waking and early wakings. Difficulty settling was determined to be due to an over-stimulating bedtime routine, and was treated with rearranging his bedtime activities so that the activities decreased in intensity as bedtime approached. Sleep interfering behaviours were identified as being maintained by parental attention and access to tangible items, which was treated by giving him access to these before bed, engaging in a bedroom tidy up routine before bed that helped to distinguish between playtime and bedtime and limited visual access to toys, as well as parents giving him limited attention through a minimal check procedure. All of the families sleep goals were obtained, with significant improvements in the children's sleep onset latency, sleep interfering behaviours and total sleep time (Jin et al., 2013).

Four studies were found that used FBA to inform their treatment plans for children with ASD who were co-sleeping. Didden et al. (2002) used FBA to inform a treatment plan for four individuals with developmental disabilities, including a 6-year-old boy with ASD, who had sleep problems that included co-sleeping, difficulties settling, and frequent night wakings that were accompanied by disruptive behaviours. A pre-treatment FBA followed ruling out any possible medical causes, a parent interview, and sleep diaries that captured the antecedents, consequences and duration of the night wakings. It was determined that parental attention acted as positive reinforcement that maintained the sleep problems in all participants (Didden et al., 2002). An extinction procedure to remove parent attention was implemented in each case, resulting in normalised sleep that was maintained at a six month follow up assessment (Didden et al., 2002).

In the study by Moore (2004), FBA was used to guide the treatment plan for a 4-year-old boy with ASD, severe learning disorders and communication delays and sleep problems that included co-sleeping, long sleep onset latencies, frequent night wakings, and early wakings. Information gained from an interview, nighttime video recordings, sleep diaries, and the Motivational Assessment Scale (Durand, 1988) lead to a hypothesis that his behaviours were being maintained through access to attention and tangible items. In addition, the PSQI identified very high stress levels in the parents, and it was therefore decided that any intervention was to be intensive and child orientated and to create as little stress as possible. A consistent bedtime routine was put in place that included a social story and visual supports. The boy was given lots of parent attention before bed in an effort to decrease his need for it at bedtime, and a

graduated extinction programme to decrease his reliance on his parents was used. Within two nights, co-sleeping was eliminated, and his sleep onset latency significantly reduced. No data was given about his night waking behaviours however. In addition, this intervention was deemed socially acceptable by his parents.

Weiskop et al. (2001) conducted a case study with a 5-year-old boy with ASD and sleep difficulties that included co-sleeping, and frequent night wakings. Through an interview and sleep diaries, it was determined that his behaviours were maintained by parent attention. Intervention involved a bedtime routine, reinforcement strategies, and a standard extinction procedure. When an extinction procedure that extinguished all parent attention at bedtime and following night wakings was introduced in the second week, positive changes were seen and he consistently fell asleep alone and remained in his own bed all night.

Weiskop et al. (2005) expanded the research by Weiskop et al. (2001) to include 13 1-9 year old children, six whom had ASD, and seven with fragile X syndrome, all with sleep problems, that included co-sleeping, difficulties settling, bedtime refusal, night wakings and early wakings. FBA was conducted through parent interviews and sleep diaries. In all cases, it was found that night wakings were maintained due to the child having learnt associations between inappropriate sleep stimuli at bedtime and sleep. When these stimuli were not present following a night waking, they needed to be reestablished to induce sleep. Disruptive settling behaviours were a result of a lack of cues that bedtime was approaching, and problematic child behaviours were positively reinforced by being allowed to co-sleep, which also negatively reinforced the parents as this

resulted in cessation of the aversive behaviours. Interventions involved bedtime routines, reinforcement strategies, and visual supports. In collaboration with the parents, all chose standard extinction over a graduated extinction procedure. Improvements were seen in the children falling asleep alone, no co-sleeping, as well as decreased pre-sleep disturbances, and night wakings. In most cases, these changes were all seen after extinction was introduced. Little evidence was given that the intervention had much impact on the children's sleep onset latencies or early morning wakings.

Summary of Functional Behaviour Assessment. FBA is a means of assessment that creates hypotheses about the antecedent and consequence factors that maintain problem behaviours, and directly guides the selection of evidence-based treatments (Beavers et al., 2013; Blampied, 2013; Brown et al., 2013; Hanley, 2016; Jin et al., 2013). Many studies have found FBA-informed interventions to greatly reduce problem behaviours in typically developing children as well as children with developmental disabilities (Beavers et al., 2013; Campbell, 2003; Hanley et al., 2003; Hanley, 2016; Horner et al., 2002). Evidence is starting to emerge that supports its value in treating sleep problems in children with ASD (Didden et al., 2002; Jin et al., 2013; Moore, 2004; Papadopoulos et al., 2015; Weiskop et al., 2001; Weiskop et al., 2005).

Research Questions

This review has demonstrated the range of interventions available for the treatment of sleep disturbance in children with ASD. The focus has been on behaviour interventions and the use of FBA to inform interventions, especially for children with sleep difficulties that include co-sleeping. In terms of

antecedent-based interventions, sleep hygiene is routinely incorporated into behaviour interventions, and is considered an essential foundation for healthy sleep (Jan et al., 2008; Mindell et al., 2009; Singh & Zimmerman, 2015; Vriend et al., 2011). Social stories and other visual supports are also used as a complementary tool to more comprehensive interventions, and help aid the child's comprehension of an intervention (Bozkurt & Vuran, 2014; Moore, 2004; Test et al., 2011). Given the difficulties that many children with ASD have with sensory modulation, and the potential impact this has on sleep problems, stimulus substitution may be an effective means to alleviate sleep problems. However, no studies were found that assessed the efficacy of this tool in the sleep literature. Faded bedtime with and without response cost, and sleep restriction hold promise as treatments for sleep problems in children with ASD, but studies investigating these procedures have lacked methodological rigor and replication (Brown et al., 2013; Vriend et al., 2011). Consequence based interventions have had more research attention, with extinction and graduated extinction procedures receiving the most empirical support for sleep problems in children with ASD (Richdale, 2013; Richdale & Wiggs, 2005; Vriend et al., 2011). Minimal check and parental presence procedures also hold promise as tools to eliminate sleep behaviours maintained by parental attention in children with ASD, but only one study for each was found that incorporated these into the interventions. Several studies have treated sleep problems using multimodal methods of intervention, with varying success (Bartlett & Beaumont, 1998; Johnson et al., 2013; Knight & Johnson, 2014; Malow et al., 2016; Montgomery et al., 2004; Papadopoulos et al., 2015; Reed et al., 2009). Combining behaviour treatments with pharmacological interventions has seen a

more rapid improvement in sleep behaviours in some cases (for example Cortesi et al., 2012; France et al., 1991; Selim et al., 2006). There is a scarcity of methodologically rigorous studies that investigate the efficacy of different treatment types on sleep problems in children with ASD (France & Blampied, 2005; Meltzer & Mindell, 2014; Turner & Johnson, 2013).

Few studies have investigated the effects of sleep interventions on co-sleeping as a target variable. Co-sleeping is not normally an isolated problem, typically co-occurring with other sleep problems, suggesting it is a complex problem with multifactorial etiologies. Given the high prevalence of reactive co-sleeping in the ASD population, more research is needed in this area.

Despite the availability of treatment options for sleep problems in children with ASD, and the complexity and heterogeneity of their abilities and sleep problems, there is a lack of studies that have used a comprehensive assessment process to inform an individualised treatment plan (Hanley et al., 2014; Jin et al., 2013). FBA is a valuable tool well suited to informing sleep interventions in children with ASD, and some studies have begun to use FBA for this purpose (Didden et al., 2002; Jin et al., 2013; Moore, 2004; Papadopoulos et al., 2015; Weiskop et al., 2001; Weiskop et al., 2005). Treatments are more effective when based on FBA than when a generic intervention is used (Beavers et al., 2013; Campbell, 2003; Hanley, 2016; Hanley, 2003; Horner et al., 2002). Co-sleeping is an example of a complex sleep problem that requires a comprehensive assessment to determine its etiology and treatment plan. FBA is lacking from the sleep intervention literature for children with ASD (Kodak & Piazza, 2008), especially with co-sleeping problems.

Studies have begun to investigate the impact of sleep on the daytime behaviours and symptomatology of children with ASD (Cohen et al., 2014; Mazurek & Sohl, 2016). These studies have demonstrated a link between sleep problems in these children and increased internalising and externalising problem behaviours, stereotyped behaviours, and social and communication deficits (Malow et al., 2006; May et al., 2015; Reed et al., 2009; Sikora & et al., 2012; Tudor et al., 2012). However, these studies are mostly cross-sectional and do not allow for an understanding into the directionality of this impact. Therefore, more research is required to ascertain the effects that successful sleep interventions have on these challenging behaviours.

Studies have also begun to look at the relationship between sleep problems in children with ASD and the families' wellbeing. Again, most of these are cross-sectional, finding negative correlations between the child's sleep and family functioning. This is another area in great need of further research.

FBA is an intervention approach that incorporates parents' preferences and goals in to the development of a treatment plan (Jin et al., 2013). In sleep interventions that are implemented by the parents in their own home, treatment needs to be a good fit for the family to increase treatment adherence and the likelihood of a positive outcome (Richdale, 2013; Turner & Johnson, 2012). Most sleep interventions with children with ASD do not report on the collaboration between the researchers and the parents, or the level of social acceptability of the treatment for the family, meaning little is known about the social validity of these interventions.

The current study aims to further the research in this area by addressing some of these challenges. The purpose of this research is to:

- 1) investigate the ability of FBA to inform intervention for sleep problems that include co-sleeping in children with ASD;
- 2) examine the effectiveness of the selected treatments on sleep-related outcomes;
- 3) evaluate the impact successful treatment has on daytime behaviours and ASD symptomatology;
- 4) evaluate the impact successful treatment has on parent well-being; and
- 5) evaluate parent acceptability and understanding of the treatment process.

Chapter 3

General Method

This thesis comprises two studies. The method described below was common to both studies. The first study is a pilot case study. The other study was a multiple baseline design across the remaining five participants.

The Sleep Research Team

The studies reported here were conducted for the researcher's thesis as part of a wider research project undertaken by a research team at the University of Canterbury.

Ethics and Participant Consent

The study received ethical approval from the University of Canterbury Human Ethics Committee (#HEC 2014/150). Parents provided written informed consent and children provided assent in keeping with their developmental levels. A copy of the child's information sheet is attached in Appendix A, and the child consent form is attached in Appendix B. The parent information sheet is attached in Appendix C, and the parent consent form is attached in Appendix D. Video consent for recording children's sleep and the recording of clinical interviews was also obtained from all families. A copy of this form is attached in Appendix E.

Participants

Recruitment. Participants were recruited throughout New Zealand through self referral or referral to the study via community service providers.

Screening and confidentiality. An initial screening process was completed over the telephone to ensure the research was appropriate for the participants, and that they met the eligibility criteria for the study. Before any screening questions were asked, the aim of the study, a basic procedural outline, and information relating to confidentiality were conveyed verbally to the family. The screening and confidentiality process took approximately 15 minutes to complete. An outline of the screening questions is included in Appendix F.

Inclusion/exclusion criteria. Children were eligible for inclusion in this study if they met the following criteria: (a) were between 2 and 12 years of age; (b) had a diagnosis of Autism Spectrum Disorder, as verified by a pediatrician, psychiatrist or registered psychologist (APA, 2013); (c) had some form of sleep disturbance which included problematic co-sleeping, as indicated by parent-report, and corroborated by sleep diaries, and (d) had limited ability to verbally communicate, defined as scores more than one year below their chronological age on the receptive and expressive communication domains of the Vineland Adaptive Behavior Scales, Second Edition, Caregiver Rating Form (VABS II; Sparrow et al., 2005). Children were excluded from the study if they had a medical condition that contributed toward their sleep problem or that impaired their ability to follow procedures in this study, or if the family was under stress in a manner which made intervention unwise.

Participant characteristics. The participants were five boys and one girl, aged between 2 years, 10 months and 6 years, 4 months. In order to maintain

participant confidentiality, pseudonyms have been assigned for each participant, and individual participant's ethnicities are not reported. Participants came from varying backgrounds, ranging from a single-parent family who did not work, through to two income families. Four of the six children were European/Pakeha, one child was Maori/Samoan, and one child was Pakeha/Indian. All children had an ASD diagnosis diagnosed by a pediatrician, with three having formal diagnoses of comorbid developmental delay. Four participants were on medication at the time the study commenced. The research team had a policy to work conjoint with medication where prescribed. A summary of participant characteristics at time of recruitment is presented in Table 1.

Table 1. Summary of Participant Characteristics

Name	Age (Y-M)	Gender	Diagnosis	Mode of diagnosis	Sleep problems	Medication
Ben	6-2	Male	ASD, Developmental Delay	Pediatrician	Co-sleeping Night awakenings	Melatonin
George	3-9	Male	ASD	Pediatrician	Co-sleeping Night awakenings	Vallergan
Harry	4-9	Male	ASD	Pediatrician	Co-sleeping Long sleep onset Night awakenings	
Catherine	4-5	Female	ASD, Developmental Delay	Pediatrician	Co-sleeping Night awakenings Early wakings	
Andrew	3-8	Male	ASD, Developmental Delay	Pediatrician	Co-sleeping Night awakenings Early wakings	Melatonin
Matt	2-10	Male	ASD	Pediatrician	Co-sleeping Night awakenings	Melatonin

Setting. Families were recruited throughout New Zealand. Depending on the location of the family, the clinical intake interview took place either in the family home or at the Pukemanu Dovedale Centre at the University of Canterbury. A researcher travelled to meet with families in their home prior to commencing intervention in order to conduct a clinical interview, administer

psychometrics, provide sleep diaries, and set up video equipment. All interventions were based in the families' homes, and implemented by the children's primary caregiver/s. Daily contact was made either through Skype, via phone, text, or email.

General Materials

Due to the uniqueness of each child's problems and intervention, materials specific to each case are described in the corresponding case methods. However, the following materials were used across multiple participants.

Video equipment. Behaviours that occurred in the child's room after they were put to bed were recorded by either a Swann-Advanced-Series DVR4-1200 camera, or a D-Link HD Cloud Camera, both of which were infrared capable and able to record for the duration of the night. The video equipment was supplied and set up by the researcher. The small video camera was placed in an elevated and inconspicuous position in the child's bedroom, pointing towards the bed. The DVR4 camera was linked to a monitor placed outside of the bedroom, enabling the parents to see the child's behaviour and to ensure the correct position of the camera, without disturbing the child. Video data for the DVR4 and DSLR cameras was recorded on to an internal hard drive and micro SD card respectively, and later uploaded on to an external hard drive for viewing. Written instructions on how to operate the equipment was provided to the families. Parents using the DVR4 camera were instructed to turn on the camera immediately before bidding the child goodnight and to turn it off upon waking in the morning. The D-Link camera was preset to automatically record from approximately ½ hour before the child's goal bedtime to 1 hour after their

expected wake time. Video recordings were used to triangulate information from the sleep diaries, provide inter-observer agreement data, and allow a more precise measurement of the child's sleep interfering behaviours. Video data was collected for a minimum of 30% of nights for each family.

Visual supports.

Social stories. Social stories were developed for each of the children included in the study. These were individualised according to the goals of intervention and the treatment procedures. The researcher determined what photos were required to include in the story, and these were collected from the parents prior to intervention. In accordance with Gray's (2010) recommendations, the text accompanying the photographs conveyed the child's new routines and were very brief, simple, and written in first person narration. These were laminated and bound together by a book ring, which allowed for the addition and subtraction of pages should modifications be needed during the intervention.

Groclocks. The Groclock™ was used by Ben, George, Harry, Andrew and Catherine to aid the understanding of the distinction between sleep and wake time. The groclock is a digital clock with glowing screens that change at preset times, to display either a sun to indicate wake time or a star to indicate sleep time.

Dependent Variables

Dependent variables were determined individually for each child, based on their presenting problems and parental goals. Each of these behaviours was

recorded from when the child was bid goodnight to when the child woke for the day. Definitions for common dependent variables are described below.

Awake. Awake was defined as any form of sleep-interfering behaviours (see below), with eyes open, vocalization of any sort, or excessive physical movement in the bed or under the covers (Jin et al., 2013).

Asleep. Asleep was defined as the child lying in bed with their eyes closed with an absence of voluntary vocalisations or movements outside of clear sleep startle and movements consistent with REM sleep.

Co-sleeping. Co-sleeping is defined as the child lying in the same bed with another individual for any period of the night, be it until they have fallen asleep, for the duration of the night, or following a night waking. This included child-initiated co-sleeping, in which the child sought out a parent and lay in their bed, as well as parent-initiated co-sleeping, in which a parent lay in the child's bed.

Parental presence. Parental presence is defined as a parent being in the presence of the child during sleep onset. This included the parent being in physical or visual proximity to the child.

Sleep-interfering behaviour. Sleep-interfering behaviours are any behaviours that occurred after the child was bid goodnight that may interfere with their ability to establish the behavioural quietude necessary to fall asleep. Such behaviours were defined as time (in minutes) spent a) vocalising (for example, singing, humming, giggling, crying, calling out, making requests, talking, or screaming); b) sitting up in bed, standing in bed, or getting out of bed; or c) engaging in stereotypic behaviours (for example, self-stimulatory behaviour

such as head shaking, hand flapping, body rocking, or repetitive manipulation of objects).

Sleep onset latency. Sleep onset latency was defined as the amount of time that elapsed between when the child was bid goodnight and the time they fell asleep.

Night waking. A night waking was defined as an arousal in which the child did not self-reinitiate sleep (Henderson, France, Owens, & Blampied, 2010; Knight & Johnson, 2014). Any waking that occurred before the time that the family deemed it appropriate for the child to wake to commence the day was considered a night waking.

Curtain calls. Curtain calls were defined as behaviours where the child remained in bed, but made requests (for example for parental attention, or access to food, drinks, toys), or when the child got out of bed and sought out parental attention or access to preferred items. Every occurrence of one of these behaviours was recorded as a curtain call.

Study Phases

The five general phases of the study are detailed below, with individual variations described under each case study.

Assessment. The assessment phase involved conducting all FBA measures, sleep outcome measures, parent-wellbeing measures and measures of the child's daytime functioning and behaviour. This information was gathered to inform the treatment plan, and to gauge any changes following treatment. Parents were advised to carry on as normal and not make any changes to their child's sleep patterns so that information would accurately reflect their situation.

Baseline. The family determined the baseline start day, to ensure that the completion of the baseline phase could be directly followed by commencement of the intervention phase. Families were advised to begin treatment on a night when they had minimal commitments the following day, in case their sleep was disrupted more than normal. During this baseline phase, parents were requested to manually complete sleep diaries and collect video recordings every night. They were instructed to not make any changes to their child's sleep routines during this time, in order for data to accurately reflect the existing sleep patterns of each child, and so that any changes in behaviour could be attributed to the introduction of the intervention, and not natural variations in behaviour (Blampied, 2013; Kazdin, 1981).

Intervention. Intervention commenced the night following conclusion of baseline. Based on assessment information the researchers developed an intervention plan that was unique to the needs of each family. The goal of treatment was to increase sleep conducive behaviours, and decrease sleep interfering behaviours. Prior to completion of the baseline phase, a meeting was held between the researchers and the family. At this meeting, the families were given the details of the proposed intervention, and were told about the relevance of the treatment to their specific problems. Families were welcomed to express their views about the acceptability of the plan for their family, and worked with the researchers to adapt any aspects deemed necessary to change. An intervention plan was agreed upon, and the families were given any resources (e.g. Gro clock, social stories) required to complete the intervention.

During the intervention, families were given support through daily phone calls or text messages with the researcher. These phone calls allowed the

researcher to document the child's sleep progress, and resolve any problems that arose as soon as possible. Daily contact meant that treatments could be promptly revised if the target behaviour did not change in the predicted way, prior to parents feeling incompetent and demoralised, therefore maximizing the likelihood of successful outcomes (Sanders & Burke, 2014).

Intervention phases varied in duration according to the needs and outcomes of each child and family, but were continued until a marked improvement in target behaviour was observed and both the researcher and family felt that the family could continue on independently.

Maintenance. During the maintenance phase, participants and researchers had no contact with each other. This phase gave the families time to consolidate the new behaviours they had acquired in to their everyday lives (Blampied, 2013; Sanders & Burke, 2014), as well as giving parents an opportunity to self-regulate and solve any problems that may have arisen without involvement of the researcher (Sanders & Burke, 2014). Post-intervention psychometrics and the post-treatment interview were conducted during the maintenance phase. The maintenance phase lasted between 4 and 6 weeks for all participants.

Follow-up. The purpose of follow-up was to measure the maintenance of treatment effects over time. Parents recorded sleep diaries for a one-week period for both follow-up periods. Video data was also recorded where possible, during this phase. Short and long-term follow-up data was gathered at six and 12 weeks post-intervention for Ben, the pilot participant. Short-term follow-up was gathered 6 to 8 weeks post intervention for George, Harry and Andrew.

Measures of Communication

The Vineland Adaptive Behaviour Scales-II, Parent/Caregiver Rating Form (VABS-II; Sparrow, Cicchetti & Balla, 2005). The VABS-II, Parent/Caregiver Rating Form is a semi-structured interview given to parents or caregivers of 0-90 year olds. It is designed to measure the individuals adaptive functioning, that is their ability to function socially and independently within their everyday environment (Gleason & Coster, 2012; Sparrow et al., 2005; Tassé et al., 2012). Subsections of the VABS-II can be administered independently from the rest of the interview (Sparrow et al., 2005). For the purpose of the current study only the communication subdomain was used. The communication domain measures an individual's level of written, receptive and expressive communication, and identifies their relative areas of strengths and weaknesses (Sparrow et al., 2005). The informant is asked to indicate how often particular behaviours are typically performed without help on a three-point scale: 2 (Usually), 1 (sometimes or partially), or 0 (never); and gives them the option of stating that they 'don't know'. Standardised scores provide an adaptive level and age equivalent.

The VABS-II is well established, with extensive normative data and strong psychometric properties (Tassé et al., 2012). It has been used considerably for children and adolescents with difficulties in intellect and independence (Gleason & Coster, 2012; Tassé et al., 2012) and is commonly used in research with ASD populations (for example, Gabriels et al., 2005; Sikora et al., 2012).

The VABS-II was used at the assessment phase to ascertain the child's level of receptive and expressive language, important for determining their eligibility for the study, and to guide the tailoring of interventions to their level of

understanding (for example, whether the use of social stories would be beneficial). It was administered by the researcher or an intern psychologist to one parent of each child.

Outcome Measures

FBA measures. The FBA process was conducted using a combination of clinical interviewing, completion of the SATT, and sleep diaries. The results of the FBA were used to develop comprehensive, individualised interventions for each child.

Clinical Interview. An open-ended clinical interview was conducted during the assessment phase. The clinical interview followed the format of a standard intake interview used by Child and Family Psychologists at the Pukemanu Dovedale Clinic, and was conducted by the researcher or an intern psychologist under supervision of a registered clinical psychologist. Consent was confirmed and confidentiality reiterated before initiating the interview. The interview was a means to collect information about the nature and history of the child's sleep problems, and the environmental conditions that envelope the child's sleep. Examples of questions used during the clinical interview are attached in Appendix H. The Sleep Assessment Treatment Tool (SATT; Jin et al., 2013) was used during the clinical interview process to guide the FBA of each child's sleep problems. The SATT explicitly features questions that investigate or identify a) the history of the child's sleep problems; b) the parents sleep goals; c) the specific sleep problems (bedtime routine noncompliance, sleep interfering behaviour, delayed sleep onset, night awakenings, early awakenings), with

descriptions of any antecedents or consequences that occur before or after the behaviour; d) the child's current sleep schedule; e) the child's pre-sleep routines; f) the child's sleep environment; g) any sleep dependencies; and h) any sleep interfering behaviours. Examples of questions derived from the SATT are included in Appendix I. The interview also provided an opportunity to determine previous attempts to alleviate the sleep problems, any possible risk factors, and any medical or physical factors that may have impacted on the child's sleep problems in the present or past. The intervention allowed for a discussion about the family's sleep related goals and gave the family an opportunity to ask any questions that they had. Meeting with the families face to face allowed for an observation of how the family members interacted, and helped the researcher and families to develop a rapport. The interview took approximately 1½ hours to complete.

Sleep diaries were also used to inform the FBA.

Sleep outcome measures.

Sleep diaries. Sleep diaries are commonly used as a method of obtaining information about children's sleep (Blampied, 2013; France & Blampied, 2005; McLay & France, 2014). In the current study, the researcher provided sleep diaries in printed form to each family, and instructed the parents on how to fill them out. The parents recorded sleep diaries for a minimum of two weeks during the assessment phase, and then every night during the baseline, intervention, and follow-up phases of the study. All families recorded information about 1) the child's daytime sleep: setting, time put to bed, time awake; 2) nighttime sleep: setting, time put to bed, the frequency, nature and parental response to curtain

calls, time until silence; 3) nighttime awakenings: time and duration of awakening, the child's behaviours while awake, and parental responses; and 4) the time that the child woke for the day. The sleep diaries were formatted so that the parents could either enter data by circling a code or writing a description of behaviours that occurred. Sleep diaries and codes were individualised for each family depending on the specific child behaviours and parental responses that were common occurrences for them. This allowed for behaviour to be quickly and consistently noted during the night by either parent. For example, the code 'LR' was used to indicate that the child left the room, and 'R' indicated the parent physically returned them to bed. A copy of a standard sleep diary is attached in Appendix G.

During the assessment phase, diaries were collected at least once per week to ensure the families were completing the diaries accurately, to inform the development of the treatment plan, and to confirm eligibility for the study. During the baseline phases, diaries were collected at least once per week. During the intervention phase, the researcher made daily contact with the families to collect sleep diaries so that necessary changes to intervention strategies could be implemented promptly. During follow-up, sleep diaries were collected on completion of the week of recording. Upon receiving sleep diaries, data was graphed and visually analysed.

Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000). The CSHQ is a 45-item parent report instrument that is used to identify and categorise behaviourally and medically based sleep problems children (Owens et al., 2000). Parents are asked to report the frequency of

particular sleep behaviours observed in their child over the previous week on a three point scale: *Usually* (5-7 nights per week), *sometimes* (2-4 nights per week), or *rarely* (0-1 night per week). In addition, they note whether these particular behaviours are a problem for the family. The CSHQ provides a total sleep disturbance score, as well as eight subscale scores relating to specific sleep disturbances most commonly observed in the pediatric population: Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night Wakings, Parasomnias, Sleep Disordered Breathing, and Daytime Sleepiness.

The CSHQ has good psychometric properties (Hodge, Parnell, Hoffman, & Sweeney, 2012; Hoffman, Sweeney, Gilliam, & Lopez-Wagner, 2006). Owens et al. (2000) found that it has adequate internal consistency for a community sample ($\alpha=.68$), as well as in a clinical sample ($\alpha=.78$), plus acceptable test-retest reliability (ranging from .62-.79). The CSHQ demonstrated validity, being able to distinguish between clinical and control groups, with a sensitivity of 0.80 and specificity of 0.72 (Owens et al., 2000).

The CSHQ is a widely used tool to study sleep in typically developing children (for example, Krakowiak et al., 2008), and is the most commonly used standardised measure of sleep problems in children with ASD (for example, Lambert et al., 2016; May et al., 2015; Mazuerk & Sohl, 2016). It has received a well-established rating from the American Psychological Association Division 54 Evidence-Based Assessment Task Force (Lewandowski, THarry-Sokol, & Palermo, 2011).

For the current study, the CSHQ was completed by one parent during the assessment and maintenance phases, to classify the nature of the sleep problem

and to determine any changes in the presentation of sleep problems following treatment.

Parent-wellbeing measures.

The Depression Anxiety and Stress Scales (DASS-21; Lovibond & Lovibond, 1995). The DASS-21 is a 21-item retrospective self-report instrument used to quantify features of depression, anxiety and stress in adults. The results of the DASS-21 provide an indication of the presence of psychological distress (Henry & Crawford, 2005). Participants are asked to rate how specific statements have applied to them over the previous week on a four point scale: *Never* (did not apply to me at all), *Sometimes* (Applied to me to some degree, or some of the time), *Often* (Applied to me a considerable degree, or a good part of the time), *Almost Always* (Applied to me very much, or most of the time). The DASS-21 provides severity labels for the depression, anxiety and stress axes, defining each as either 'normal', 'mild', 'moderate', 'severe', or 'extremely severe'.

The DASS-21 has good psychometric properties (Henry & Crawford, 2005). Henry and Crawford (2005) found that it has adequate reliability ($\alpha=.82-.90$ for the subscales), and good convergent and discriminative validity. The DASS-21 is a widely used tool to assess adult well-being in both clinical and research settings, and has been used extensively in research with parents of children with ASD (for example, Giallo et al., 2011).

For the current study, the DASS-21 was administered to both parents during assessment and maintenance phases, to assess any changes in levels of parental wellbeing. The DASS-21 form took parents approximately five minutes to complete.

Treatment acceptability measures.

Post-treatment interview. In order to gain an understanding of the parents perspective of the treatment process, a semi-structured interview was held with parents during the maintenance phase. During the interview parents were asked how they felt about the intervention, and the overall process. They were asked their perspectives of how and why the treatment was effective, and whether the outcome or process had any impact on them personally, or on other areas of their child's behaviour or development. Finally, they were asked for any suggestions as to how the process could be improved. An outline of the post treatment interview questions is included in Appendix J. This interview took approximately ten minutes to complete.

Treatment Acceptability Rating Form – Revised (TARF-R; Reimers & Wacker, 1992). The TARF-R is a 20-item parent-report questionnaire used to measure the acceptability of treatments for children in naturalistic settings (Reimers & Wacker, 1992). 17 of the items measure the interventions acceptability, by asking parents to consider how appropriate, effective and fair they deemed it to be. The remaining three items assess the parent's perception of the severity of their child's behaviours, and their understanding of the treatment. Through the use of a 7-point Likert scale, items are summed to give a total acceptability score, with higher responses indicating a more acceptable treatment. The TARF-R has good reliability ($\alpha=.92$) and clinical utility (Finn & Sladeczek, 2001; Reimers & Wacker, 1992). It has been used with a variety of populations, including research settings evaluating treatments for challenging

behaviours in children with autism (for example, Lee, Anderson, & Moore, 2014; McLay, Carnett, van der Meer, & Lang, 2015).

For the current study, the TARF-R was administered to parents during the maintenance phase. Used in conjunction with a semi-structured interview, the researcher was able to gauge the parent's acceptance of the treatment, with the additional advantage of receiving feedback that could improve any future research.

Inter-Observer Agreement. Inter-observer agreement (IOA) was obtained by comparing sleep diaries recorded by the family with the video recordings. A research assistant blind to the sleep diaries viewed at least 20% of the video recordings across each phase of the study. IOA data was recorded on to the same sleep diary template that was used by each family. Frequency of behaviours, i.e., number of curtain calls and frequency of night wakings, was recorded as agreement if both the parent and observer noted the behaviours occurrence, and disagreement if only one party noted the behaviour. Measures of duration, i.e. length of sleep onset latency and duration of night wakings, were recorded as agreement if parent and observer reports were within 15 minutes of each other. Percentage of agreement for each behaviour was calculated using the equation $[\text{Agreements}/(\text{Agreements} + \text{Disagreements})] \times 100$.

Chapter 4

Pilot study

Ben

Ben was a 6 year, 4 month old boy who had been diagnosed by a pediatrician as having ASD and Global Developmental Delay when he was 4 years old. Ben had limited verbal language skills, communicating using one to two word utterances. Ben received an age-equivalent score of 1 year, 6 months and 1 year, 8 months on the receptive and expressive subdomains of the VABS –II (Sparrow et al., 2005) respectively. Ben lived at home with his parents, younger brother, and intermittently with his grandfather. He attended a local inclusive primary school. Ben was prescribed melatonin in order to reduce his sleep onset latency. Parents had reported that this was effective and were continuing to use it.

Presenting Complaints

Ben's parents made contact with the researchers due to their concerns regarding his inability to settle to sleep independently in his own bed, and the occurrence of co-sleeping upon night wakings. Ben's parents reported that on a typical night, he would indicate to them that he was tired by taking himself to his choice of bed. This was typically his parents' or grandfather's bed. He would ask for the lights to be turned off. On most occasions, his parent would bid him good night and leave the room. Ben would then proceed to get out of bed between one

and three times, being returned to bed each time, before his father would eventually lie with him until he fell asleep. If he started to sleep in his parents' bed, they would transfer him to his own bed during the night without waking him. If he woke during the night and no one was with him, he would leave the room and seek out his parents or grandfather to sleep with. If he woke and someone was there, he would resume sleep. When he co-slept, Ben liked to be in full body contact with his bed partner, and would tuck his legs under them. Approximately once a week, Ben would have an hour-long day nap, typically in the car.

Co-sleeping with family members had occurred since birth. This behaviour had been maintained over time as the family felt that it ensured that Ben would get sufficient quantity and quality of sleep. The family also felt that they would be better able to ensure Ben's safety while they slept as he had a tendency to wander around the house.

Previous attempts to eliminate co-sleeping included staying in the same room with him on an adjacent mattress. However, this was unsuccessful, as Ben would simply move to the other mattress with his father. Ben's parents had not persisted with any one strategy for a prolonged period of time as they felt that the strategies were ineffective.

Ben's parent's goals were for him to 1) settle to sleep independently in his own bed during sleep onset, and 2) to remain asleep in his own bed for the duration of the night (i.e. without co-sleeping). Criteria for meeting these goals required complete absence of his parents during these periods of sleep initiation or reinitiation, and that he must fall asleep in his own bed.

Functional Behaviour Assessment

Results of the FBA indicated that Ben's sleep difficulties were maintained by multiple possible factors. A varied sleep location and a dependence on the sensory input from physical contact during sleep onset were interfering with his ability to independently settle to sleep. His inability to self-settle meant that when he woke in the night and the same contingencies were not in place, he was unable to resume sleep. FBA also indicated that Ben received positive reinforcement in the form of a family members attention during night wakings. As a result, it was hypothesised that the primary functions of his sleep disturbances were social attention and to gain sensory stimulation.

Method

Research design. An AB case study design was used.

Materials specific to Ben.

The 'body'. This 'body' was designed to replicate the warmth and size of another person and acted as a replacement for the presence of a parent. The 'body' was a standard maternity pillow, with dimensions of 140cm (W) x 48 cm (H) x 20cm (D). The original pillow was filled with polyester, and covered in non-woven polypropylene. The pillow was modified by covering it in Minion™ patterned material, and adding a pocket to each end of the pillow allowing for the insertion of hot water bottles. These hot water bottles added warmth and weight to the 'body', and were heated or not at the parents' discretion.

Baseline. Ben's baseline phase ran for a period of 28 days, until a stable pattern of data emerged. Data is missing for 2 nights of baseline as the family was not home and diaries were not recorded.

Intervention.

Treatment Phase One. Phase One of treatment started on Day 29, and included elimination of daytime naps, video modeling, extinction of parental presence during sleep onset, the introduction of a sleep item, and a reinforcement procedure.

Elimination of daytime naps. In an effort to promote good sleep hygiene and apply sleep pressure, Ben's parents were asked to eliminate daytime naps. This was achieved by directing Ben into sleep incompatible activities (e.g., giving him a drink, or engaging him in an activity) if he showed signs of tiredness.

Video modeling. A short 32-second iMovie was created for Ben to reflect the changes in his sleep routine, and to help him understand the expectations around sleep. The video portrayed Ben getting ready for bed, and sleeping the entire night in his own bed, without a parent or grandparent present. Steps in the movie included Ben having a bath, putting on his pyjamas, eating his dinner, relaxing with his family, going to bed with the 'body', cuddling it if he woke during the night, and getting a reward if he slept all night alone in his own bed. His parents voiced dialogue from Ben's perspective that accompanied video footage, for example "At night I go to sleep with my Minions" and "If I wake up at night I cuddle my Minions and go back to sleep". The video was shown to Ben at least once every night at the start of his bedtime routine, and during the day upon his request.

Extinction of parental presence during sleep onset. During sleep onset, Ben's parents guided him to his own bed, bid him goodnight, and then left the room. They then ignored all curtain calls. If Ben left his room, they would return him, help him to restore the sleep position, and direct him to the 'body' without engaging in any additional verbal or physical interactions. They then left the room again.

Introduction of a sleep item. Ben was provided with the 'body' (see materials section). This was given to him when up was put to bed each night. He was able to cuddle into the 'body' during sleep onset and upon night wakings.

Reinforcement procedure. Ben was rewarded with access to movies, contingent upon him sleeping alone in his own bed all night.

Procedural modifications. The treatment plan was altered during the course of intervention, resulting in four main treatment phases. A breakdown of each additional treatment phase is as follows:

Treatment Phase Two.

Extinction of co-sleeping during the night. On Day 30, a procedure was introduced to eliminate co-sleeping during the night. The initial plan was to focus on eliminating co-sleeping during sleep onset first, to determine if this was enough to impact on his sleep behaviour during the night. However, Ben's parents decided to start this earlier. All other treatment procedures remained constant. As with sleep onset, the parents immediately returned Ben to bed if he attempted to co-sleep during the night. Any other attentions seeking behaviours (e.g., calling out to his parents) was also ignored.

Treatment Phase Three.

Groclock. On Day 45, a Groclock was introduced in an attempt to teach Ben the time that he was able to get up in the morning. This was set, so that the star changed to a sun at 6am. Ben was instructed that if the Groclock had a picture of a sun on it, he could exchange it for the iPad in the morning. He was not given access to the iPad until the sun was on the clock.

Treatment Phase Four. On Day 49, visual signs were introduced, and the iMovie updated to reflect the changes.

Visual signs. Laminated pictures of the star and sun that were on the Groclock were tacked to Ben's parents and grandfathers doors. They changed these visuals at 6am, consistent with the Groclock. Ben was instructed that if he woke in the night, and saw the star sign on the doors, he was to return to his bed and resume sleep. By contrast, if he saw the sun, he was able to go in and see his parents and request the iPad by handing over the sun symbol.

iMovie. Scenes were added to the iMovie to reflect the changes to the use of visual stimuli. For example, movie clips synchronized with the words "If I wake up during the night and I see a star on my clock, I cuddle my Minions and go back to sleep" or " When I see a sun on Mum and Dad's or Grandad's door I can go in and seen them".

Follow-up. Short and long term follow up data was collected for one week each, starting 41 and 83 days following the completion of intervention.

Results

Ben's treatment lasted for a period of 161 days. Data is missing for nights 120 and 156 to 164, as the family was holidaying and hence they were not in their normal environment.

Sleep outcomes.

Sleep diaries.

Effect on sleep onset latency. Ben's data for duration of sleep onset latency (SOL) and frequency of curtain calls (CC's) are represented in Figure 1.

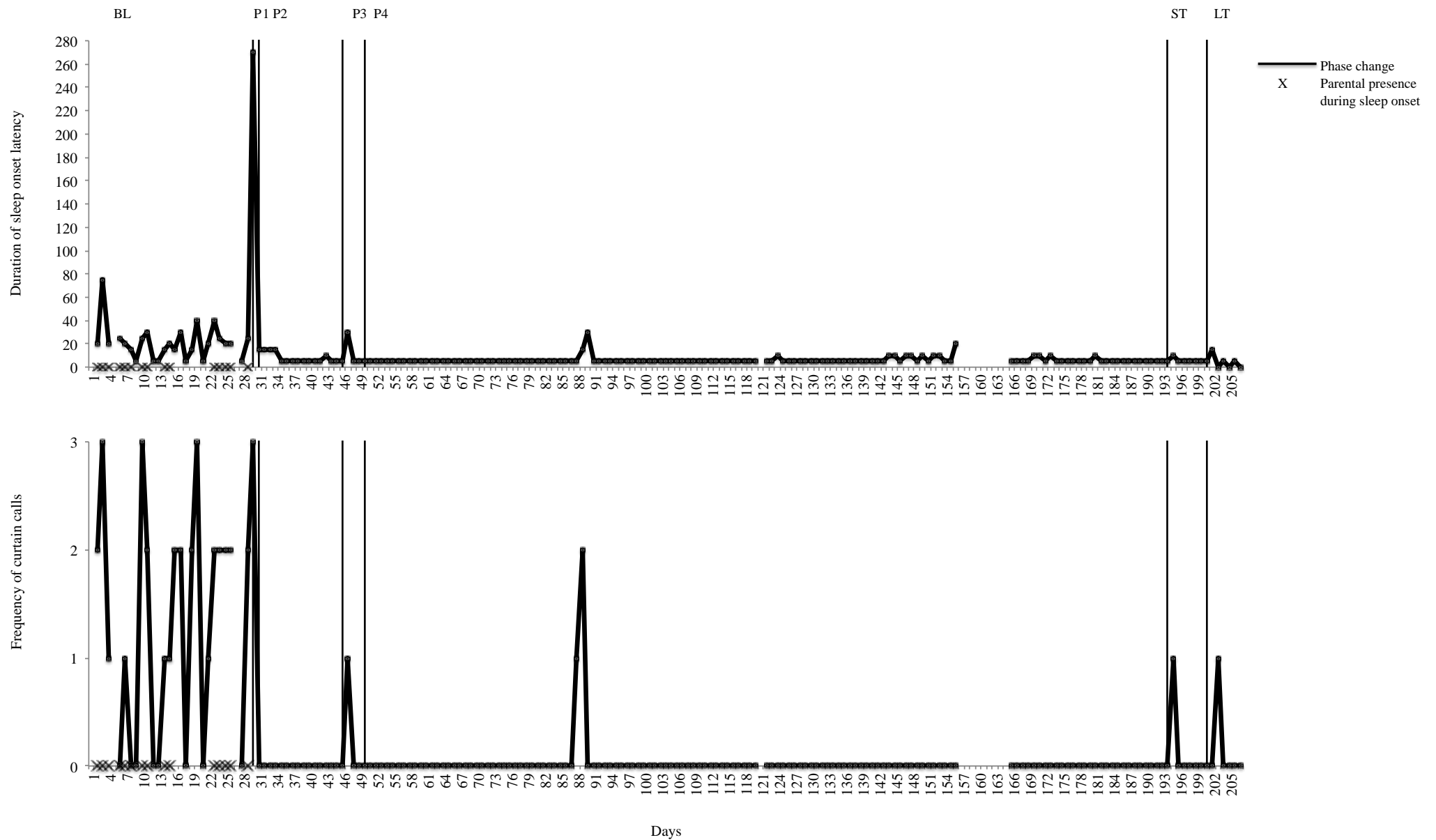


Figure 1. Duration of sleep onset latency and frequency of curtain calls across baseline, intervention and follow up phases for Ben.

For Ben, his SOL was variable during baseline (between 5 and 75 minutes). After an initial escalation of his SOL to 270 minutes on the first night of intervention, his SOL rapidly declined to 15 minutes on the second night of intervention. Thereafter, SOL was less variable than baseline, and typically lasted for 5 minutes. At short-term follow-up, SOL remained short (5-10 minutes). This represents a marked reduction from baseline. This was also maintained during long-term follow-up, with a SOL of 1-15 minutes.

Effect of curtain calls. During baseline, the frequency of CC's varied between 0 and 3. On the first day of intervention, Ben had three CC's. This decreased to zero on day two. Zero CC's were observed during the remainder of the intervention, with the exception of the 46th, 87th and 88th nights of intervention where between one and two CC's were observed. Looking in detail at the sleep diaries, Ben had a day nap on all three of these days, which was out of character for him during the intervention phase. These naps may have decreased his sleep pressure for those nights. On each occasion, Ben was vocalizing, and then left his room, whereupon his father returned him to his own bed. Ben's parents reported a great reduction in the frequency of curtain calls during follow-up, with one CC during both the short- and long-term follow-up periods, and the remaining nights with zero CC's.

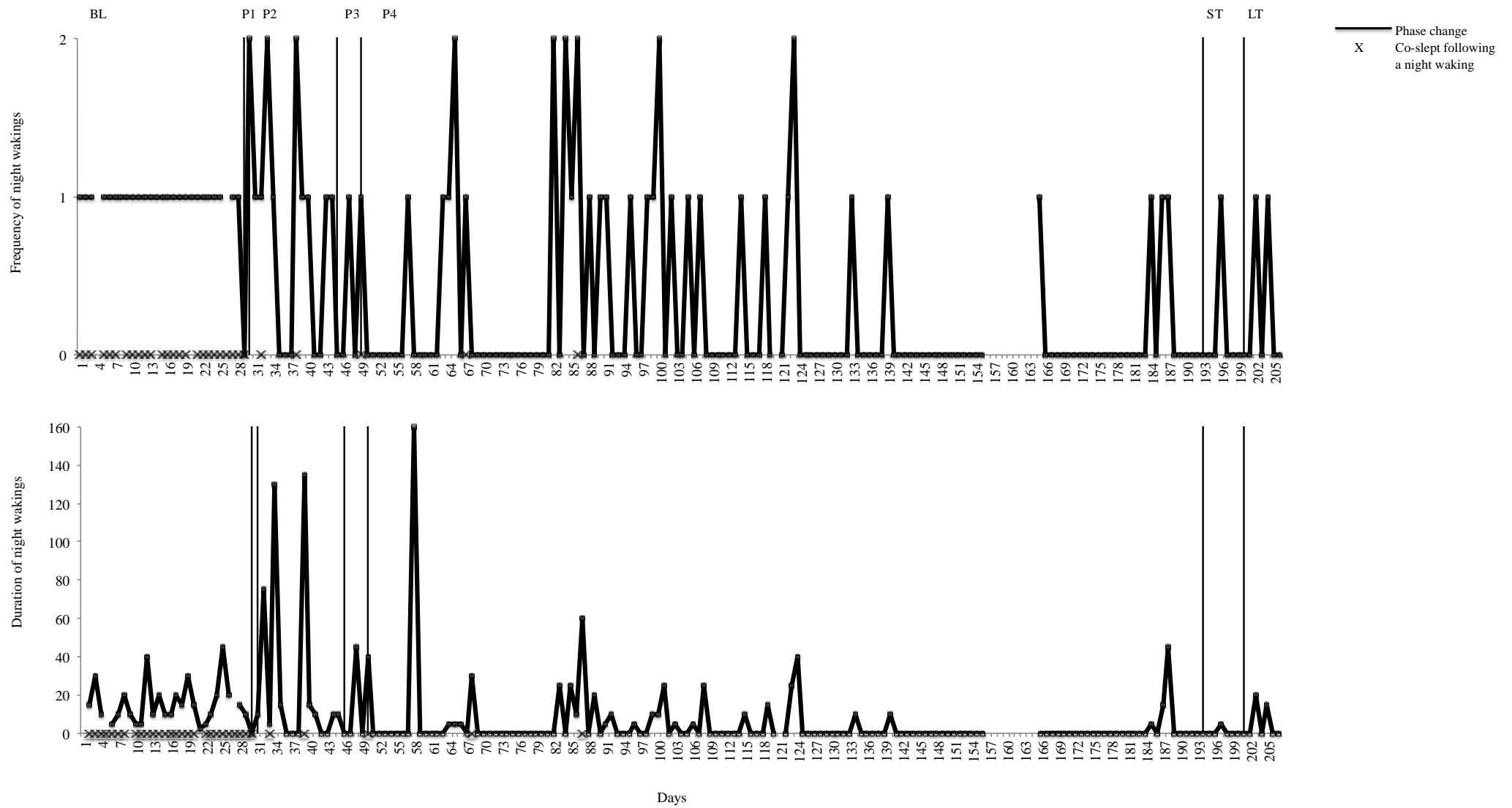


Figure 2. Frequency of night wakings and duration of night wakings across baseline, intervention and follow up phases for Ben.

Effect on night waking. The frequency and duration of night wakings (NW's) are represented in Figure 2. During baseline, Ben consistently woke one time every night. This was typically followed by the occurrence of co-sleeping. Ben had no NW's on the first night of intervention, but this escalated to two NW's with the introduction of phase 2 (extinction of co-sleeping during the night). From the first day of intervention until day 140, Ben's frequency of NW's became variable, with 1-2 NW's occurring for 37 out of 111 nights. However, he also began to have several nights where he did not wake and seek attention at all. From day 124 to day 183 of the intervention phase, Ben had no NW's for the majority of nights, with only 1 NW per night on three occasions. On days 133 and 139, Ben left his room, and was returned to bed by his father, being awake for just 10 minutes each time. Day 165 was Ben's first day back after an overseas holiday. He had had a nap on the plane, which was likely to decrease his sleep pressure that night, and it is also possible that he was readjusting to time zone changes. There was some variability in frequency of NW's in the follow-up phases, however marked improvements were seen compared to baseline. At short-term follow-up, Ben did not wake 6/7 nights. At long-term follow-up, Ben did not wake 5/7 nights. The pattern of night waking duration mirrored that of frequency, with longer durations typically observed on nights where Ben woke more frequently.

Effect on Sleep Goals. Progress towards the families' goals is represented in Figure 3. This goal chart conveys the extent to which the families sleep goals were achieved. Depicted measures for Ben include parental presence to initiate sleep and co-sleeping during the night. Filled squares represent nights during

which a particular sleep goal was attained, and open squares represent nights during which a particular sleep goal was not attained. (NB: Due to the duration of the intervention, a sample of every fourth week was graphed). During baseline, 31% of goals were met on recorded nights. Ben's parents' goal of settling to sleep independently during sleep onset was consistently met from the first day of intervention, and was maintained during both follow-up phases. With the exception of five days, co-sleeping following a night waking was eliminated from the first day that extinction was implemented. At the start of intervention, on days 32 and 38, Ben crept in to bed with his grandfather, who did not return him to bed. On day 49 and 86, Ben co-slept with his father after very long night wakings, and on day 67, he co-slept after being very distressed from a nightmare. These results demonstrate that with intervention, Ben was able to consistently settle to sleep independently in his own bed, and was able to self-settle during the night. These results were maintained long term.

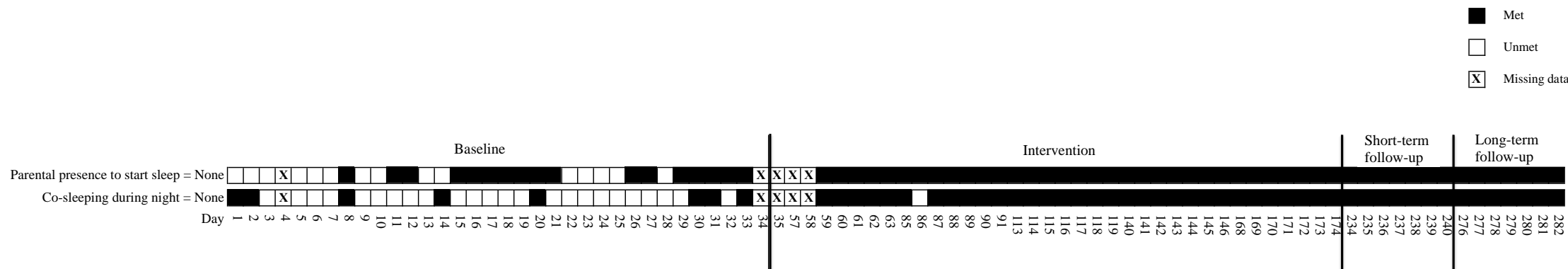


Figure 3. Goal attainment across baseline, intervention and follow-up phases for Ben.

The Child Sleep Habits Questionnaire, abbreviated form (CSHQ: Owens et al., 2000). Results of the CSHQ (Owens et al., 2000) are presented in Table 2. The higher the score, the more a problem is indicated. The CSHQ results show that Ben's parents reported a reduction in bedtime resistance from 13/24 to 9/24 between pre- and post-intervention. Sleep onset delay and night awakenings remained consistent.

Table 2. Comparison of Pre- and Post-Intervention Scores on the CSHQ Completed by Ben's Parents

Variable scores	Pre-intervention	Post-intervention	Maximum score
Bedtime resistance	13	9	24
Sleep onset delay	2	2	4
Night time awakenings	6	6	12

Effect on parents wellbeing.

DASS-21. Results of the DASS-21 (Lovibond & Lovibond, 1995) are presented in Table 3. A higher score indicates an increased risk of disturbance. The DASS-21 suggested that both Ben's mother and father had normal levels of depression, anxiety, and stress throughout their participation in the study. However, both parents had a minimal increase in anxiety ratings, and marked decreases in stress levels between pre-and post-intervention.

Table 3. Comparison of Pre-and Post-Intervention Scores on the DASS-21 for Ben's Mother and Father

Variable scores	Mother		Father	
	Pre-intervention	Post-intervention	Pre-intervention	Post-intervention
Depression	0	0	0	0
Anxiety	0	1	0	1
Stress	5	0	6	1

Treatment acceptability.

Treatment Acceptability Rating Form – Revised (TARF-R). Table 4

displays the results of the TARF-R (Reimers et al., 1992a) for each of Ben's parents pre- and post-intervention.

Table 4. Post-Intervention Treatment Acceptability Scores from TARF-R for Ben's Parents.

Variable scores	Mother	Father	Maximum score
Total acceptability	90	84	119
Reasonableness	20	19	21
Willingness	15	18	21
Cost	10	10	14
Side-effects	14	11	21
Effectiveness	19	15	21
Disruption/Time	12	11	21
Problem severity ^o	12	12	14
Understanding of treatment ^o	6	7	7

^o = not included in the Total Acceptability Score

Overall, results showed a satisfactory level of treatment acceptability, with scores of 90/119 and 84/119, for Ben's mother and father respectively. Results suggest that both parents felt the treatment was reasonable given the types of problems (mother: 20/21, father 19/21). The TARF-R scores also suggest that Ben's father was more willing to carry out the intervention than his mother (18/21 and 15/21 respectively), but his mother thought the treatment was more effective in resolving Ben's sleep issues than his father (19/21 and 15/21 respectively). Results suggested that both parents felt that much time was needed to carry out the intervention (mother:12/21, father:11/21), that there was a real possibility of undesirable side-effects (mother:14/21, father:11/21), and there was some cost involved with the treatment (both:10/14). Ben's parents indicated that they understood the treatment process to a high degree (mother: 6/7, father: 7/7).

Post treatment discussion. Ben's mother stated that they were very satisfied with the overall treatment process and that the treatment effects had exceeded their expectations. She said that it was initially very hard to implement the intervention strategies as they were so tired themselves, but that the psycho-education helped them to understand the need to be consistent, and the improvements they saw gave them the motivation to continue. She believed that sharing and alternating the intervention demands between the two parents, as well as psycho-educating the extended family, was vital to the treatments success. She said she felt that the 'body' had the most impact on Ben's sleep behaviour improvements. She said that she felt that the intervention provided them with the self-efficacy and confidence to resolve any sleep related issues that may arise in the future. She said that the treatment has also had a significant impact on the rest

of the family, with both parents “feeling human again” due to getting more sleep themselves and getting some routines set in place. She also said that as a result of the intervention, Ben’s brother was getting more sleep. In addition, Ben’s mother reported improvements at school, which she contributed to him getting more sleep. Since he began the sleep intervention, she believes his attention improved, with him being able to sit at a desk and being able to attend to tasks for long periods of time. He also engaged in writing in school, showing more perseverance in this area.

Inter Observer Agreement. IOA was unable to be coded. A video camera was initially installed in Ben’s bedroom, but was removed at the parents’ request, due to the noise of the camera disrupting his sleep.

Chapter 5

Study 2

Method

After the pilot participant completed intervention, the study procedures and measures used were adapted to enhance the experimental design and methodology, and to further explore the collateral effects of improved sleep outcomes on children's daytime functioning, ASD symptomatology, and parental wellbeing. The Questions About Behavioral Function (QABF; Matson & Vollmer, 1995) was introduced in order to support the FBA process. The Child Behavior Checklist (1½-5 years) (CBCL (1½-5); Achenbach & Rescorla, 2000) and Gilliam Autism Rating Scale, Third Edition (GARS-3; Gilliam, 2016) were introduced as measures of the impact of sleep intervention on the child's daytime behaviours and ASD symptomatology, and the Relationship Quality Index (RQI; Norton, 1983) and Pittsburgh Sleep Quality Index (PSQI; Bursse, Reynolds, Monk, Bermna, & Kupfer, 1989) were introduced to capture more information regarding the impact of a sleep intervention on parental relationships and sleep quality.

Research Design

A single-case A-B multiple baseline across participants design was used for the five participants. Single-case designs draw inferences about individuals, through the repeated gathering of core dependent variables within and across different conditions or phases (Blampied, 2014). A single-case design was chosen because of the idiographic approach to the research. Although all children

partaking in this study shared an ASD diagnosis and co-sleeping problems, they varied in regard to ASD symptomatology, co-occurring sleep problems, and the variables that were maintaining the sleep problems. A single-case design allowed for the complexities associated with this heterogeneous population to be retained and factored in to all aspects of assessment, treatment, and data analysis. Single-case studies fit with a scientist-practitioner model allowing for research to be carried out in clinical settings, and preventing individuals from waiting in a 'control group' before receiving intervention (Blampied, 2013; Spruyt & Curfs, 2015).

A multiple baseline across participants design allows for the evaluation of behaviour change across multiple participants with similar behaviours (Blampied, 2014). Interventions are introduced at varying time points, permitting the analysis of change in behaviour as a direct result of intervention. Being able to aggregate individual cases together provided potential for stronger evidence of treatment effects when compared to information gained from a single case.

Multiple sources and methods of data collection were gathered to triangulate findings and counter any biases that may arise as a result of the flexibility granted to researchers in a case study design (Vertue, 2011). The use of a multiple baseline, as well as continuous sampling across multiple time points, and a concerted effort to account for alternative and competing explanations for any behavioural changes, helped to minimise the impact that confounding variables may have had on the internal validity of the study (Kazdin, 1981; Vertue, 2011).

Baseline. Families were randomly assigned to a one, two, three or four week baseline phase, using the True Random Number Generator (Random.Org).

Outcome Measures

The psychometrics that were added to Study 2 are described below.

Functional Behaviour Assessment Measures.

Questions About Behavioral Function (QABF; Matson & Vollmer, 1995).

The QABF is a 25-item form that is used to generate hypotheses about the function/s of the target behaviour (Freeman, Walker, & Kaufman, 2007; Healy, Brett, & Leader, 2013; Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2000).

Research suggests that treatments that are informed by the results of the QABF are more effective in reducing challenging behaviours than those that are not (Freeman et al., 2007; Paclawskyj et al., 2000). The QABF consists of five subscales (Social Attention, Escape, Non-social Reinforcement, Physical Discomfort, and Tangible Reinforcement), which include five items each. The items are scored on a four-point scale: *Doesn't apply* (Never), 1 (Rarely), 2 (Some), and 3 (Often).

Number of items and severity ratings are summed separately for each subscale.

Ranking each subscale determines what the caregiver believes is the most prominent function or functions of the behaviour. The QABF is currently the functional assessment scale with the strongest psychometric properties (Matson, Tureck, & Rieske, 2012). It has good test-retest reliability, moderate to good inter-rater reliability, and good internal consistency (Freeman et al., 2007; Healy et al., 2013; Matson et al., 2012; Paclawskyj et al., 2000).

The QABF was completed by one parent during baseline.

Measures of daytime functioning.

Child Behavior Checklist (1½- 5 years) (CBCL (1½-5); Achenbach & Rescorla, 2000). The CBCL (1½-5) is a 100-item standardised parent report measure that assesses internalising and externalising behaviours in preschoolers aged 18 months to 5 years. The parent is asked to report the frequency of behaviours that they observe in their children on a three point Likert scale: 0 (not true), 1 (somewhat or sometimes true), or 2 (very true or often true). Scores are summed and converted to T scores, with higher scores indicating greater behaviour problems across seven syndrome scales (emotionally reactive, anxious/depressed, somatic complaints, withdrawn, sleep problems, attention problems, and aggressive behaviour), and five DSM-5-oriented scales (depression, anxiety, ASD, ADHD, and oppositional defiance). Syndrome scales are combined to form internalising problems, externalising problems and total problems composite scores. T scores are used to determine whether the child's score represents normal, borderline, or clinical behaviour.

Considerable reliability and validity data has been published for the CBCL (Iyanova et al., 2010). The CBCL has been used extensively in research investigating the link between ASD, sleep problems and challenging daytime behaviours (for example, Anders, Iosig, Schwichtenberg, Tang, & Goodlin-Jones, 2012; Delahaye et al., 2014; Fadini et al., 2015; Goldman et al., 2009; Hollway, Aman, & Butter, 2013; Lambert et al., 2016; Moon et al., 2011; Sikora, Johnson, Clemons, & Katz, 2012). An intern psychologist or the researcher administered the CBCL during the baseline and maintenance phases.

Gilliam Autism Rating Scale, Third Edition (GARS-3; Gilliam, 2016). The GARS-3 is a 58-item professional or parent report screening instrument used to assess the probability of ASD and the level of symptom severity. It is based on the diagnostic criteria of the DSM-V (APA, 2013), and can be used for individuals aged 3-22 years. Informants are asked to rate items on how adequately they describe the individual's behaviour on a four-point scale: 0 (Not at all like the individual), 1 (Not much like the individual), 2 (Somewhat like the individual), or 3 (Very much like the individual). Six subscale scores related to difficulties most often observed in individuals with ASD (restrictive/repetitive behaviours, social interaction, social communication, emotional responses, cognitive style, maladaptive speech) are combined to yield an Autism Index score that assesses the probability of ASD and the degree of severity.

The GARS-3 is normed on children and young adults diagnosed with ASD, and has good psychometric properties (Gilliam, 2016). It is a consistent and discriminative tool, with fair to excellent internal consistency (α scores ranged from .79 to .94 for the subscales), good test-retest reliability, and strong inter-rater reliability (Gilliam, 2016).

The GARS-3 was administered during baseline and maintenance phases.

Parent well-being measures.

Relationship Quality Index (RQI; Norton, 1983). The RQI is a six-item self-report questionnaire given to couples to assess their perceptions of their relationship quality and satisfaction (Sanders, Markie-Dadds, & Turner, 2001). Participants individually rate the extent to which they agree with statements about their relationships on a 7 point Likert scale (1= very strongly disagree through to

7= very strongly agree). The scores are summed to indicate global relationship satisfaction, with the higher scores representing greater satisfaction. The RQI was completed by both parents separately during assessment and maintenance to monitor any changes in relationship quality.

Pittsburgh Sleep Quality Index (PSQI; Bursse, Reynolds, Monk, Berman, & Kupfer, 1989). The PSQI is an 18 item self-report measure used to evaluate quality of sleep in adult populations (Bursee et al., 1989). Participants are asked questions relating to their usual sleep habits over the past month. 14 of the 18 questions use a four point Likert-type scale to assess the frequency of particular sleep behaviours: 0 (not during the past month), 1 (less than once a week), 2 (once or twice a week), or 3 (three or more times a week), as well as seven subscale scores relating to sleep quality, sleep onset, actual sleep, sleep efficiency, sleep disturbance, sleep medication, and daytime sleepiness. The PSQI provides a global score with higher scores denoting more impaired sleep.

Bursse et al. (1989) reported good psychometric properties for the PSQI, with an internal reliability of .83, and test-retest reliability of 0.85. The global score correctly identified 88.5% of participants, and had a sensitivity of 89.6% and specificity of 86.5%. The PSQI has been used in both clinical and research settings, and it is commonly used in research to measure the sleep quality of parents and caregivers of autistic children (for example Giallo, Wood, Jellet, & Porter, 2011; Hodge et al., 2013; Hoffman et al., 2008; Lopez-Wagner et al., 2008; Meltzer, 2008). The PSQI was administered to both parents during assessment and maintenance, to assess any changes in parents' sleep quality over the course of treatment.

Data Analysis

Data obtained through sleep diaries during the baseline, intervention, and follow-up phases were graphed according to the common dependent variables across participants. This included duration of sleep onset latency, frequency of curtain calls, frequency of night wakings, and duration of night wakings. For Andrew, additional graphs that displayed the number of times he was returned to bed at different stages of the night were also analysed. The primary means of data analysis was systematic visual inspection of the graphed data, comparing the study phases within each case, and across all participants. Visual analysis is a powerful way to assess treatment outcomes, and is commonly used with single-case multiple baseline across participant designs, as it enables one to ascertain whether behaviour changes are attributable to treatment or not (Blampied, 2013; Hanley et al., 2003). Visual analysis of graphs included assessment of the mean, level, trend, variability, latency and consistency of sleep behaviours (Cohen, Feinstein, Masuda, & Vowles, 2014).

Parental goals for their child's sleep were plotted on a chart. Goal attainment could be inspected visually to detect any changes between and within study phases.

Outcomes of pre- and post-treatment measures for the CSHQ, DASS-21, PSQI, RQI, GARS-3, and CBCL were analysed within and between participants before and after treatment, to detect any changes as a result of the study.

Participants

Matt

Matt was a 2 year, 10 month old boy who had been diagnosed with ASD by a pediatrician when he was 2 years, 2 months old. Matt would communicate using one to two word utterances. Matt received an age-equivalent score of 1 year, 2 months and 1 year, 9 months on the receptive and expressive subdomains of the VABS-II respectively. Matt lived at home with his mother, father, and six siblings that ranged in age from 10 months to 17 years.

Presenting Complaints

Matt's mother made contact with the researchers due to her concerns regarding his inability to initiate sleep independently, co-sleeping following night wakings, and early morning wakings. Matt was given 3mg of melatonin each night immediately prior to bedtime. Matt had a consistent bedtime routine and was put to bed by 7pm each night. His mother would then sit beside his bed until he fell asleep. This typically took 5-20 minutes. If she left before he was asleep, he would become distressed, and would leave the room to look for her. Matt would typically wake at about 2am, and would climb in to his parent's bed. If he became distressed, they would give him another bottle of milk with melatonin, and it would take him about 1 ½ hours to return to sleep. During this time he would fidget and bounce on the bed. He would typically wake for the day at about 6am. Matt shared a bunk bed with an older brother who went to bed after Matt was asleep.

Matt's mother reported that since he was about 2 years, 8 months old, he had developed a need to know where his mother was at all times during the day,

becoming quite distressed when he could not find her. She further reported that his sleep behaviours had always been difficult. Until he was 18 months old, he would wake every 45 minutes, needing parental assistance to get back to sleep. After his younger brother was born, he had a period of good sleep that lasted for about 2 months. However, following this, Matt would take 3 to 4 hours to get to sleep, and would require his parents presence to reinitiate sleep following a night waking. When he was 2 years, 4 months old they moved him from a cot to a single bed, and started giving him 1mg melatonin, but they reported that this had little impact on his sleep.

Matt's parents had tried several techniques to decrease his sleep onset period and need for parental presence. This included driving in the car, elimination of daytime naps, and waking him in the night to provide additional melatonin. When Matt turned 2 years, 7 months of age, his dose of melatonin was increased to 3 mgs, which had a positive effect on his sleep onset latency.

Matt's parents goals were for him to: (1) settle to sleep independently during sleep onset and night wakings, (2) to remain asleep in his own bed for the duration of the night, and (3) wake after 6am. The criteria for meeting Goal 1 was the complete absence of his parents during sleep onset, and without a parent lying with him, following a night waking. The criterion for Goal 2 was that he did not seek out his parents during the night, and the criterion for Goal 3 was that he did not wake for the day until after 6am.

Functional Behaviour Assessment

Results from the FBA indicated that dependence on parental presence during sleep onset, co-sleeping, night wakings, and early wakings was interfering

with Matt's ability to independently settle to sleep. His inability to self-settle meant that when he woke in the night and the same contingencies were not in place, he was unable to resume sleep. FBA also indicated that Matt received positive reinforcement in the form of parent attention during night wakings. It was hypothesised that the primary function of his sleep disturbance was parental attention. This was confirmed by the QABF results.

Method

Baseline (BL). Matt was randomly assigned a one-week baseline phase.

Intervention.

Treatment Phase One (P1). Phase one of treatment included the elimination of daytime naps, introduction of cuddle time prior to bedtime, faded bed time, a parental presence procedure, a reinforcement procedure, and a social story.

Elimination of day sleeps. In an effort to promote good sleep hygiene and apply sleep pressure, Matt's parents were asked to eliminate daytime sleeps. Should they notice that he was starting to fall asleep during the day, they were asked to redirect him into engaging activities that promoted wakefulness (e.g., distract giving him a drink, or engaging him in a game or conversation).

Introduction of allocated cuddle time. In an effort to provide social attention that would not interfere with sleep onset, Matt's mother was asked to incorporate special cuddle time on the couch as a part of the bedtime routine.

Delayed bedtime. In order to apply sleep pressure, Matt's bedtime was moved from 7pm to 8pm.

Camping out. Matt's parents were instructed to begin a camping out procedure. Initially, his parents were to sit on the floor next to Matt's bed until he fell asleep. They were asked to avoid providing any physical or verbal attention unless deemed necessary for his safety. If Matt attempted to leave his bed, his parents were asked to return him to bed and restore the sleep position without providing verbal or social attention. The distance between Matt's parents and his bed was to be gradually increased, until parental presence was no longer required for him to fall asleep. For example, his parents were to begin by sitting on the floor next to his bed, then move to the door, then move to the hallway, and then move out of sight. Movement through each step in this procedure was contingent upon improvements in Matts' sleep onset latency and frequency of curtain calls, and was done in consultation with the researcher. Following a night waking, Matt's parents were asked to respond in the same way as they did during sleep onset. Should Matt wake before 6am, his mother was to treat this as a night waking.

Reinforcement procedure. Matt received a small chocolate and verbal praise upon waking in the morning. This reinforcement was delivered contingent on him sleeping all night in his own bed, regardless of whether he needed to be returned to bed or not.

Social Story. A social story was created to reflect the changes in the sleep routine, and expectations. Photos and text depicted Matt putting on his pyjamas, having cuddle time with his mother on the couch, cuddling a toy while trying to get to sleep, his mother staying in his room until he was asleep, his mother returning to her own bed, his mother returning him to bed should he leave his room during the night, cuddling a toy while trying to get to sleep, and getting a reward if he

slept all night alone in his own bed. The story was read to Matt at least once every night during his bedtime routine, and during the day upon request.

Procedural modifications. The treatment plan was altered during the course of intervention, resulting in four main treatment phases. Each additional treatment phase is described below:

Treatment Phase Two (P2). On Day 20, the family decided to return Matt's bedtime to 7pm. They believed that he was too tired, as they felt that his daytime behaviour and language had deteriorated.

Treatment Phase Three (P3). On Day 57, the family chose to change Matt's bedtime to 8.30pm due to the heat at their holiday destination making it hard for Matt to fall asleep earlier.

Treatment Phase Four (P4). On Day 72, the family bought Matt a new bed. While on holiday, he had slept well on a larger single bed, and they wanted to try this at home.

Andrew

Andrew was a 3 year, 8 month old boy who had been diagnosed by a developmental pediatrician as having ASD and Global Developmental Delay, when he was 2 years old. Andrew had limited verbal language skills. His communication consisted mostly of echolalia and scripting of single words. Andrew received an age-equivalent score of 1 year, 1 month and 1 year, 2 months on the receptive and expressive domains of the VABS-II. Andrew lived at home with his parents and older sister. Andrew was prescribed melatonin, which they were continuing to use on occasion, in response to night wakings.

Presenting Complaints

Andrew's parents contacted the researchers due to concerns regarding the frequency and duration of night wakings, co-sleeping and early morning wakings. Andrew's parents reported that on a typical night, he would usually take himself to bed between 7 and 7.30pm. Most nights, his mother would tuck him in to bed and sit with him until sleep onset. He would typically fall asleep within 20 minutes. Andrew woke between one and three times per night, upon which he would leave his room and seek out his mother. If he woke before 4am, he required his mother to lie or sit with him in his bed, and it would take 30 minutes to 2 hours to return to sleep. Most mornings, he would wake between 4 and 5.30am. His mother would try to return him to his own bed, but his vocal and motor stereotypy, and shouting out would escalate if he remained in his room. In response to this, his mother would then take him to the lounge where he engaged in preferred activities (e.g., watching t.v.).

Andrew's sleep behaviours became a problem for the family when he transitioned from a cot to a bed, and learnt how to open a door. At this point Andrew began leaving his room to seek out his mother. At 3 years, 3 months old, he was prescribed 1 mg of melatonin. His parents reported that this was somewhat effective, and that they were using melatonin infrequently and sometimes reactively in response to night wakings or delayed sleep onset.

As well as melatonin, previous attempts to alleviate his sleep problems included using a Groclock, and holding the door so he could not leave the room, however his parents reported that these were not effective. They also tried pushing his sleep time back in an effort to lengthen his morning wake time, but reported that this was only effective for a short while.

Andrew's parents goals for him were: (1) to settle to sleep independently following a night waking, and (2) to sleep in his own bed until 6am. The criterion for meeting Goal 1 was that Matt must fall asleep in his own bed, without a parent lying with him, following a night waking. The criterion for Goal 2 was that he did not wake for the day until after 6am.

Functional Behaviour Assessment

Results from the FBA indicated that Andrew's sleep difficulties included co-sleeping, frequent and prolonged night awakenings, and early awakenings. These were maintained by multiple variables. During night wakings and early morning awakenings, Andrew received parental attention, which appeared to be positively reinforcing Andrew's sleep interfering behaviour. During early morning wakings, Andrew's early morning wakings, also seemed to be reinforced with access to preferred activities. As a result, it was hypothesised that the primary function of Andrew's sleep interfering behaviour was social attention, and the secondary function was access to tangible items. This was confirmed by the QABF results, which indicated that the primary function was attention.

Method

Baseline (BL). Andrew was assigned a 2-week baseline phase.

Intervention.

Treatment Phase one (P1). Phase One of treatment included faded bedtime, use of visual supports, camping out, a Grocloc, reinforcement, and a social story.

Faded bedtime. Andrew's bedtime was delayed by 1 hour so that he was put to bed at 8.20pm. This time was selected as sleep onset typically occurred within 15 minutes of this time. Delaying his bedtime was also introduced to create sleep pressure (i.e., to increase Andrew's biological need for sleep).

Visual support. To help Andrew to understand the time that he was able to leave his room in the morning, his door was to be fully opened and the hallway light turned on at 6am. Any waking before this time was to be treated as a night waking.

Camping out. During sleep onset and night wakings Andrew's mother implemented a camping out procedure, following the identical procedure used by Matt, except that Andrew's parents began by sitting on his bed.

Groclock. In an attempt to teach Andrew the time that he was able to get out of bed in the morning, a Groclock was used. This was set so that the star changed to a sun at 6am.

Reinforcement procedure. Andrew received a small treat and verbal praise upon waking in the morning, contingent on him sleeping independently in his own bed (regardless of attempts to co-sleep), and staying in his room past 6am.

Social story. A social story was developed that depicted Andrew putting his pyjamas on, brushing his teeth, getting in to bed, Andrew sleeping, staying in bed if he woke during the night, getting up when the hallway light was on, and getting a reward. The story was read to Andrew at least once every night during his bedtime routine, and during the day upon request.

Procedural modifications. The treatment plan was altered during the course of intervention, resulting in three main treatment phases. Each additional treatment phase is described below:

Treatment Phase Two (P2). By Day 62, it was noted that Andrew's wake times had not changed significantly. An additional 30 minutes was added to his bedtime, in the hope of applying greater sleep pressure.

Treatment Phase Three (P3). On Day 116, Andrew's parents chose to buy Andrew a slightly bigger bed, hoping that this would positively change his sleep behaviours.

Follow-up (FU). Short term follow up data was collected for one week, starting 5 weeks following the completion of intervention.

George

George was a 3 year, 9 month old boy who had been diagnosed by a pediatrician as having ASD by when he was 2 ½ years old. George had limited verbal language skills, communicating through using simple 2-3 word utterances. George had an age-equivalent score of 1 year, 6 months and 2 years, 1 month on the receptive and expressive subdomains of the VABS-II respectively. George lived at home with his mother and two older sisters.

Presenting Complaints

George's mother made contact with the researchers due to her concerns regarding his sleep patterns, specifically sleep onset delay, co-sleeping during sleep onset and night wakings, frequent and prolonged night wakings, and early

morning awakenings. George had a consistent bedtime routine, which included his mother lying with him until he fell asleep. This typically took 10 minutes. During sleep onset, George liked to link fingers with his mother, and required his hands to be squeezed. He also liked to flick his mother's hands, and enjoyed having her hands resting on his face and over his head. George would wake three to four times during the night. When he woke, George would call out for his mother and occasionally request a bottle of milk. His mother would lie in bed with him, in his bed, until sleep resumed. If she was already asleep, he would join her in her bed for the remainder of the night. George would wake for the day between 4 and 6am.

George was able to self-settle to sleep between 6 months and 1 year of age. However, his mother believed that he required her presence to fall asleep after he got croup at 1 year. George's mother had tried several strategies to address his co-sleeping and night wakings. This included the use of 30mg/5ml Vallergan Forte, prescribed by his pediatrician when he was 2 years, 7 months. This was still being used on occasion. She had also used a weighted blanket, and melatonin but she believed it made no difference to his bedtime behaviours.

George's mother's goals for him were to 1) settle to sleep without Vallergan, 2) settle to sleep independently in his own bed during sleep onset and following night wakings, and 3) to sleep in his own bed until 6am. The criterion for meeting Goal 1 was that he did not have any Vallergan before sleep onset. The criteria for Goal 2 was the complete absence of his mother from his room during sleep onset, and that Matt must fall asleep in his own bed, without a parent lying with him, following a night waking. Goal 3 was met if he did not wake for the day until after 6am.

Functional Behaviour Assessment

Results from the FBA indicated that George's sleep difficulties included sleep interfering behaviours, delayed sleep onset, night awakenings, co-sleeping and early awakenings, that were maintained by multiple possible factors. A dependence on the sensory input from his mother's physical contact during sleep onset was interfering with his ability to independently settle to sleep. His inability to self-settle meant that when he woke in the night and the same contingencies were not in place, he was unable to resume sleep. FBA also indicated that George received positive reinforcement in the form of parent attention during night wakings, and gained a tangible positive reinforcement on occasion, by being given a bottle of milk. As a result, it was hypothesised that the primary function of his sleep disturbance was social attention, the secondary function was access to a tangible item, and the third function was to gain sensory stimulation. This was confirmed by the QABF results.

Method

Materials specific to George.

Sensory Ball. The sensory ball was a 'Transparent Yuk-E-Ball™', which was a small transparent ball that contained multiple smaller balls.

Baseline (BL). George was assigned a three-week baseline phase.

Intervention.

Treatment Phase One (P1). Phase One of treatment included the introduction of allocated cuddle time, a camping out procedure, Vallergran, a groclock, a reinforcement procedure, and a social story.

Introduction of allocated cuddle time. In order to provide an appropriate opportunity for social attention, that would not interfere with sleep onset, George's mother was to incorporate special cuddle time on the couch in to the bedtime routine.

Camping out. George's mother was instructed to begin a 'camping out' procedure. This adhered to the same procedure used by Andrew, except that his mother was instructed to move further from the bed every third night in accordance to the Vallergran dosage regime (see Appendix K), as opposed to his sleep patterns.

Vallergran. To decrease any possible resistance and distress during sleep onset and night wakings, Vallergran was prescribed by George's General Practitioner, and the fading regime was approved by him and his mother. Vallergran was to be given to George in a bottle of milk every night, approximately 20 minutes before his expected sleep onset. He was to start with 1.8mls, and this dose was decreased by approximately 1/5 every three nights, until its use was extinguished (see Appendix K). This was done in conjunction with George's mother fading her presence out of his bedroom (i.e., Vallergran dose decreased on the night following his mother moving further from the bed).

Groclock. In an attempt to teach George the time that he was able to get out of bed in the morning, a Groclock was used. This was set so that the star changed to a sun at 6am.

Reinforcement procedure. George received a marshmallow and verbal praise as soon as he woke in the morning, contingent on him sleeping all night in his own bed, regardless of attempts to co-sleep.

Social Story. A short social story was created for George that depicted him putting on his pyjamas, having cuddle time with his mother on the couch, reading a book in bed with his mother, cuddling his dog, and squeezing his stress ball, his mother staying in his room until he was asleep, George sleeping, what to do if he woke during the night, using the Groclock to know when he could get out of bed, and getting a reward if he slept all night alone in his own bed. The story was read to George at least once every night during his bedtime routine, and during the day upon request.

Procedural modifications. The treatment plan was adjusted during the course of intervention, resulting in five main treatment phases. The additional phases are as follows:

Treatment Phase Two (P2). On Day 25, a stress ball was introduced as a replacement for holding hands with his mother. This was unavailable in phase one.

Treatment Phase Three (P3). By Day 35, George was requesting milk every night that he woke. An extinction programme, removing milk, was put in place and his bottle was given prior to bedtime, rather in bed. A bottle of water was available beside his bed for if he was genuinely thirsty.

Change to Vallergan and camping out regime. Due to the expectation of a PERB following the elimination of bottles, Vallergan was increased to 1.8mls, and George's mothers chair moved to within 1 metre of his bed. Adjustments made to Vallergan dosage and mothers position were thereafter contingent on

improvements in his sleep onset latency, frequency of curtain calls, and frequency and duration of night wakings, rather than a strict regime.

Treatment Phase Four (P4). On Day 75, George's mother had moved the chair so that she was sitting outside of his room, and out of view. In an effort to decrease the likelihood of a PERB resulting from this change, George's Vallergan was increased to 1.4mls. George's mother was also advised to close George's door after he was put to bed, and not re-enter unless George's safety was compromised. This was done in order to reduce the frequency with which George was leaving the room, and eliminate the reinforcing effect of returning George to bed.

Treatment Phase Five (P5). On Day 77, George's mother raised concerns about having George's door being completely closed, as he could not open it. It was decided that the door being closed would be contingent on George leaving his bedroom during sleep onset or a night waking (i.e., if he attempted to leave the room, he would be returned to a sleeping position, and his door closed). George's mother did not actually fully close his door, but put the door in a position that appeared closed from George's bed.

Follow-up (FU). Short-term follow up data was collected for one week, starting 4 weeks following the completion of intervention.

Harry

Harry was a 4 year, 9 month old boy who had been diagnosed with ASD by a pediatrician when he was 2 ½ years old. Harry would communicate using five to six word utterances. Harry received age-equivalent scores of 2 years, 2 months and

2 years, 1 month on the receptive and expressive subdomains of the VABS-II respectively. Harry lived at home with his mother, father and younger brother.

Presenting Complaints

Harry's parents made contact with the researchers due to Harry's delayed sleep onset latency, frequent and prolonged night wakings, and co-sleeping. Harry had a consistent nighttime routine, that included being in bed at 8pm. Harry would take one to four hours to settle, during which time he would talk to himself, play with his toys, and leave his bed seeking out his parents and/or to use the toilet. Harry consistently woke every night between 2 and 4am, and would co-sleep in his parent's bed. Harry usually woke for the day between 8 and 9am.

Harry was able to settle to sleep quickly and independently until he was 4 years of age. His parents were uncertain about the cause of the changes in his sleep patterns. Harry had never slept through the night, and had always woke around midnight, sought out his parents and climbed in to their bed. Harry's parents felt that he experienced separation anxiety when left alone in his bedroom.

Harry's parents had tried several strategies to address his sleep problems. This included talking in the lounge next to his room to decrease any separation anxiety, playing soft music, and a parental presence programme. They persisted with these strategies for 3-4 weeks, but felt that they were ineffective, with the exception of the parental presence programme which they felt was effective for the period in which it was implemented.

Harry's parents' goals were for him to 1) have a sleep onset period of less than 30 minutes, 2) to settle to sleep independently following a night waking, and 3) to remain in his own bed for the duration of the night. The criterion for meeting

Goal 1 required Harry to be asleep within 30 minutes of his parents bidding him goodnight and leaving his room. The criterion for meeting Goal 2 required Harry to not fall asleep in his parents bed, or a parent not sleeping in his bed, following a night waking. To meet Goal 3, Harry was not to disturb his parents during the night.

Functional Behaviour Assessment

Results from the FBA indicated that Harry's sleep difficulties included sleep interfering behaviors, delayed sleep onset, co-sleeping and night awakenings, which were maintained by multiple possible factors. Harry played in his room at bedtime to gain stimulation from playing alone in a repetitive manner, and to avoid sleep. This, as well as an entrained late sleep time, and a possible genuine need to go to the toilet, interfered with his ability to fall asleep quickly. FBA also indicated that hearing his parents talking interfered with his ability to independently fall asleep. His inability to self-settle meant that when he woke in the night and the same contingencies were not in place, he was unable to resume sleep. Also, Harry received positive reinforcement in the form of parent attention during night wakings. It was hypothesised that the primary function of Harry's night wakings and co-sleeping was parent attention for leaving his room. The main functions of his prolonged sleep onset appeared to be to gain access to toys, avoid going to bed, and a lack of sleep pressure. This was confirmed by the QABF results.

Method

Materials specific to Harry.

Finish Box. The 'finish box' was a large, clear plastic storage container with a 'finished' symbol on the top.

Music. Whale sounds were played to Harry throughout the night, using an iPad.

Baseline (BL). Harry was assigned a four-week baseline phase.

Intervention.

Treatment Phase One (P1). The treatment plan included sleep hygiene practices, music, a faded bedtime routine, extinction, a consistent wake time, a Grocloc, a reinforcement procedure, and a social story.

Sleep hygiene. In an effort to decrease noise during bedtime, Harry was moved to a bedroom in the house that was not adjacent to the lounge. Harry's parents were instructed to take him to the toilet as part of the bedtime routine, decreasing the likelihood that he genuinely required the toilet during sleep onset. As part of the bedtime routine, Harry was to pack up his toys, and put them in the 'finished box', indicating to him that the toys were not accessible at bedtime. Harry was provided access to these toys upon waking in the morning.

Music. Soothing music was played quietly to Harry throughout the night, and turned off when he woke in the morning. This was used to create non-social setting events during sleep onset that were consistent throughout the night.

Faded bedtime. Harry's bedtime was delayed until 9:45pm as this was the time at which sleep onset was likely to occur within 15 minutes. This bedtime was to be faded forward in 15 minute increments to an earlier time of 8.45, dependent on Harry consistently reaching his goal sleep onset period of less than 30 minutes.

Removal of parental attention. Due to parental preference, an extinction procedure was used during sleep onset and night wakings. After bidding Harry goodnight, his parents were instructed to give him no additional verbal or physical attention for the remainder of the night, unless deemed necessary for his safety. They were instructed to ignore all curtain calls, and should he leave the room, they were asked to return him to bed, restore the sleep position, then leave the room.

Consistent wake time and Groclock. During baseline, Harry woke between 6.30 and 10.30am. In order to establish consistent sleep patterns, apply sleep pressure, and condense sleep, Harry's parents were instructed to wake him by 7am if he was not already awake. Any wakings before 6am were to be treated as night wakings. In an attempt to increase Harry's understanding of the time that he was able to get out of bed in the morning, a Groclock was used. This was set so that the star changed to a sun at 6am.

Reinforcement procedures. Upon waking, Harry was immediately provided with access to the toys in his finished box. He received a star on a sticker chart if he slept all night alone in his own bed, regardless of whether he got out of bed during the night. Harry's parents chose a tangible reward to be given to Harry after receiving five stars on his chart.

Social story. A social story was created for Harry to help explain the new expectations around sleep. Photos and text depicted Harry in his new bedroom, putting his toys away, having a drink, having a bath and brushing his teeth, putting on his pyjamas, sleeping in his own bed all night, using the Groclock to determine when it was time to get up, playing with his toys in the morning, his sticker chart, and Harry getting a reward. The story was read to Harry at least once every night during his bedtime routine, and during the day upon request.

Procedural Modifications. From Day 46, the decision was made to provide Harry with a M&M lolly each morning contingent upon him sleeping all night alone in his own bed. Immediate reinforcement was provided as the star chart did not appear to be reinforcing for Harry.

Follow-up (FU). Short-term follow up data was collected for one week starting five weeks following the completion of intervention.

Catherine

Catherine was a 4 year, 5 month old girl who had been diagnosed with ASD and Developmental Delay by a pediatrician just after she turned 2. Catherine communicated using one to two word utterances. Catherine received age-equivalent scores of 1 year, 7 months and 2 years, 1 month on the receptive and expressive subdomains of the VABS-II respectively. Catherine lived at home with her mother, father and younger brother.

Presenting Complaints

Catherine's mother made contact with the researchers due to her concerns regarding co-sleeping, night wakings, and early morning awakenings. Catherine had a fairly consistent bedtime routine, which would start at 7pm. Catherine would have a bottle of milk in bed, and her mother would sit on the bed with her until she fell asleep. This typically took 5 minutes. During this time, her mother would whisper softly to her. About once per week, Catherine would have a day nap. On the nights that this occurred, she would go to bed later and require a parent to lie with her to fall asleep. Catherine would typically wake between two and six times

per night, leaving her room to seek out her parents. If her parents were awake, they would return her to bed and sit with her, and she would typically reinitiate sleep within 5-10 minutes. If her parents were in bed, she would ask for a bottle of milk, which she was occasionally given, and she would then co-sleep with them in their bed. Catherine would typically wake for the day before 6am.

Catherine's mother reported that she had always had difficulties settling to sleep. As a baby, her parents would get her to sleep by walking her in a pram or driving her in the car. When Catherine was about 3 ½ years old, she would wake during the night and often become upset.

Catherine's parents had tried several strategies to combat the night wakings, co-sleeping and early wakings. They had tried a reinforcement procedure and a groclock, but felt that both of these techniques were too complex for Catherine to understand at the time, and therefore stopped using them. They had also moved Catherine in to her own bedroom, and had changed her bed, but this did not improve her sleep.

Catherine's parents goals were for her to 1) settle to sleep independently during sleep onset and night wakings, 2) settle to sleep without a bottle, and 3) to wake up after 6am. The criterion for meeting Goal 1 was the complete absence of parents during sleep onset, and without a parent lying with her following a night waking. To achieve goal 2, Catherine was to not have a bottle in bed during sleep onset. The criterion for Goal 3 was that Catherine must not to get up for the day until after 6am.

Functional Behaviour Assessment

Information from the FBA indicated that dependence on her mothers' presence as well as her bottle were interfering with her ability to independently settle to sleep. Her inability to self-settle meant that when she woke in the night and the same contingencies were not in place, she was unable to resume sleep. In addition, FBA indicated that Catherine received positive reinforcement in the form of parent attention during night wakings. Based on this information, it was hypothesised that the primary factor maintaining her sleep difficulties was parental attention, a secondary function was to gain stimulation through her mothers soft talking, and a third function was to gain access to her bottle. This was confirmed by the results of the QABF.

Method

Materials specific to Catherine.

Music. Classical music was played to Catherine during the night, via an iPad.

Baseline (BL). Catherine was assigned a two-week baseline phase.

Intervention.

Treatment Phase One (P1). Phase One of treatment included the removal of daytime naps, alterations to bedtime routine, music, camping out, a Groclock, reinforcement procedure, and a social story.

Removal of day sleeps. In an effort to promote good sleep hygiene and apply sleep pressure, Catherine's parents were asked to eliminate daytime naps. Should

they notice that Catherine was starting to fall asleep during the day, they were to distract her using reasonable means (e.g., engaging her in a game or conversation).

Introduction of allocated cuddle and milk time. In order to provide an appropriate opportunity for social attention and access to her milk that would not interfere with sleep onset, Catherine's mother was to incorporate special cuddle and milk time on the couch in to the bedtime routine.

Music. Music was to be played to Catherine throughout the night to create a non-social association between the sounds and sleep.

Camping out. Catherine's parents were instructed to begin a 'camping out' procedure that was identical to that used by Matt.

Groclock. In an attempt to teach Catherine about the time that she was able to get out of bed in the morning, a Groclock was used. This was set so that the star changed to a sun at 6am. Catherine's comprehension had progressed sufficiently from the families last attempt at using a Groclock that it was considered feasible to try again.

Reinforcement procedure. Catherine received a reward (5 minutes on the iPhone), as well as verbal praise immediately upon waking in the morning. This was delivered contingent on her sleeping all night in her own bed, regardless of whether co-sleeping occurred.

Social story. A social story was provided for Catherine to reflect the changes in her sleep routine and to help her understand the expectations around sleep. The story portrayed Catherine putting on her pyjamas, brushing her teeth, being read a story, having milk on the couch, sleeping in her own bed all night, listening to music, using the Groclock to determine when it was time to get up, and getting a

reward. The story was read to Catherine at least once every night during the bedtime routine, and during the day upon request.

Procedural modifications. The treatment plan was adjusted during the course of intervention, and a breakdown of each additional treatment phase is as follows:

Treatment Phase Two. By Day 20 Catherine was refusing having her bottle before bed, therefore her mother made the decision to eliminate this from the bedtime routine.

Treatment Phase Three.

Vallergan. To decrease any possible resistance and distress during sleep onset and night wakings, Vallergan was introduced. Catherine's General Practitioner prescribed Vallergan. Vallergan was to be given to Catherine in a bottle of milk every night, approximately 20 minutes before her expected sleep onset. Catherine was to start on 0.8mls, decreasing dosage by approximately 1/5th every time, contingent on improvements in Catherine's sleep onset latency and curtain calls. Changes were made in liaison with the researcher.

Chapter 6

Study 2

Results

Chapter 6 presents data on the child's sleep outcomes as a result of sleep interventions for the Study 2 participants. In addition, data is presented that compares both parent well-being and child daytime functioning pre- and post-intervention for these families. Treatment acceptability data collected post-intervention is also reported.

Two of the 5 Study 2 participants did not complete the intervention. Matt's family discontinued their involvement with the study before he had completed intervention. At submission of the current study, Catherine had not yet completed her intervention, and was still involved with the research project. All data that was collected before intervention, as well as sleep outcome data collected throughout their interventions is presented in the results.

Summary of Participants Treatment Phases

Matt.

Treatment Phase One (P1): Elimination of day sleeps, introduction of allocated cuddle time, delayed bedtime, camping out, reinforcement, social story.

Treatment Phase Two (P2): Bedtime moved earlier.

Treatment Phase Three (P3): Bedtime moved later.

Treatment Phase Four (P4): New bed.

Campout procedure changes: Fading of Matt's mothers' presence occurred on days 14, 18, 41, 42 and 61.

Catherine.

Treatment Phase One (P1): Elimination of day sleeps, alterations to the bedtime routine, stimulus substitution, camping out, Groclock, reinforcement, social story.

Treatment Phase Two (P2): Bottle removed from bedtime routine.

Treatment Phase Three (P3): Vallergran.

Campout procedure changes: Fading of Catherine's mothers' presence occurred on days 39, 64, 68, 78, and 87.

Vallergran changes: Vallergran dosage decreased on days 94, 97 and 99.

Andrew.

Treatment Phase One (P1): Faded bedtime, visual support, camping out, Groclock, reinforcement, social story.

Treatment Phase Two (P2): Bedtime moved later.

Treatment Phase Three (P3): New bed.

Campout procedure changes: Fading of Andrew's mothers' presence occurred on days 49, 55 and 66.

George.

Treatment Phase One (P1): Allocated cuddle time, campout procedure, faded Vallergran dosage, Groclock, reinforcement, social story.

Treatment Phase Two (P2): Stimulus substitution.

Treatment Phase Three (P3): Milk removal.

Treatment Phase Four (P4): Door closed.

Treatment Phase Five (P5): Door contingencies, removal of parent presence.

Campout procedure changes: Fading of George's mothers' presence occurred on days 25, 28, 31, and 34. Her position was moved closer on day 35, and then faded again on days 40, 43, 47, 54, 60, 71, and 75.

Vallergan changes: Vallergan dosage decreased on days 26, 29, and 32. Vallergan dosage was increased on day 35, and then faded again on days 41, 45, 48, 55, 61, and 72. Dosage increased again on day 75, and then faded on days 81, 85, 87 and 92.

Harry.

Treatment Phase One (P1): Sleep hygiene, stimulus substitution, faded bedtime, removal of parent attention, consistent wake time, Grocloclock, reinforcement, social story.

Faded bedtime changes: Harry's bedtime was faded later on days 44, 51, 54, and 59.

Sleep Outcome Measures

Sleep diaries. Duration of sleep interventions varied between children, as length was contingent on the child's progress. Matt's parents did not implement the treatment programme between days 27 and 36 due to his mothers' absence from the home, and illness. He was involved in the intervention for a total of 70

days before his family chose to stop their involvement with the research due to tiredness. Data is reported for all days of Matt's intervention involvement.

At the time of submission of this thesis, Catherine had not completed treatment. Data is included for 101 days. Data is missing for days 31, 36-45, 52-59 and 67-68, as the family were on holiday.

Intervention lasted 108, 92, and 42 days for Andrew, George and Harry respectively. For Andrew, data is missing for Days 100-113 because Andrew was away from home on a holiday, and the parents did not record diaries during this time. For George, sleep diary data is missing for nine of the baseline nights, as the sleep diaries were lost.

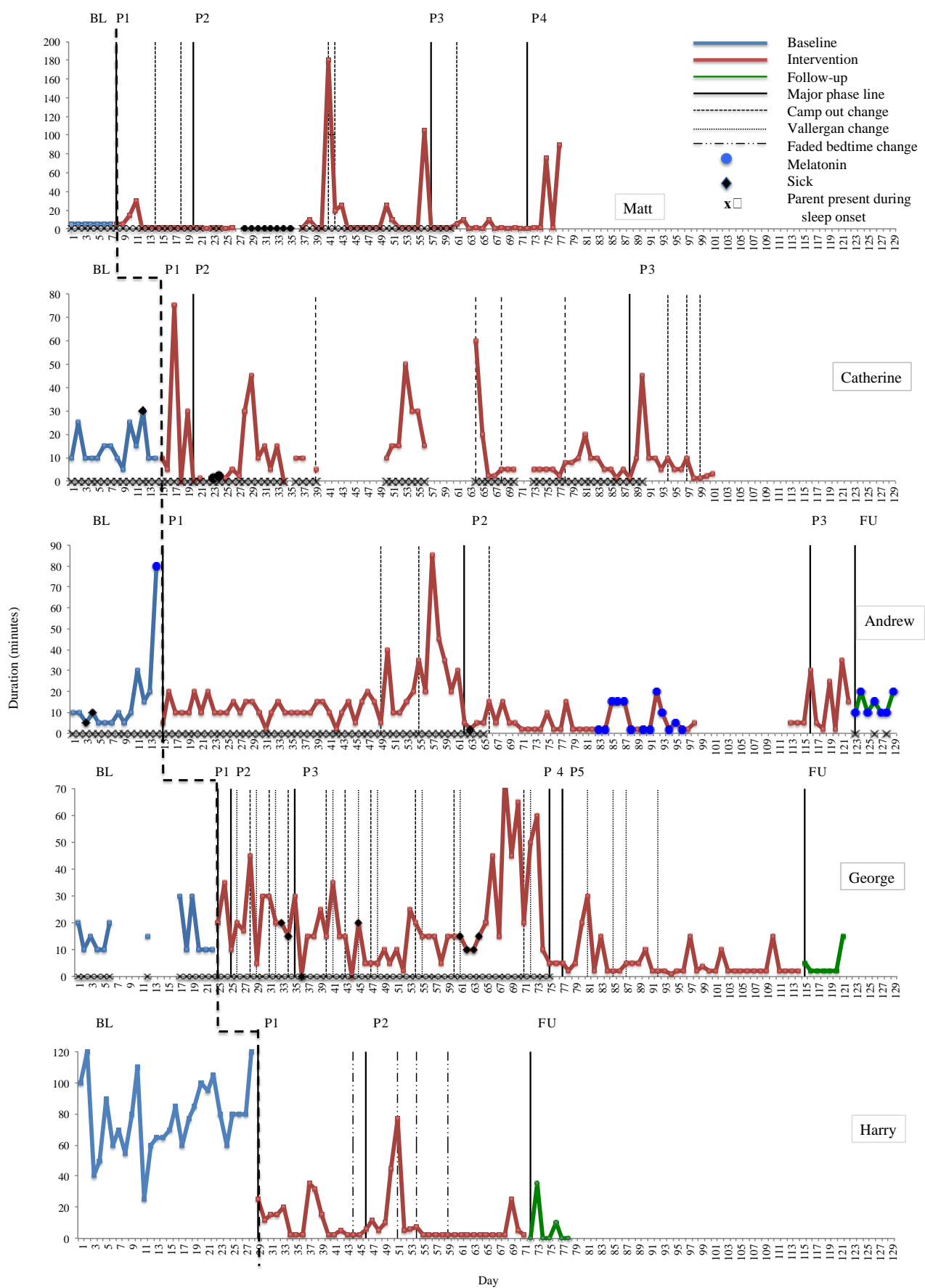


Figure 4. Duration of sleep onset latency during baseline, intervention and follow-up for Study 2 participants.

Effect on sleep onset latency. Figure 4 presents baseline, intervention and follow-up data for sleep-onset latency (SOL) for each of the five children included in this study. During baseline, SOL was highly variable for Catherine, Andrew, George and Harry. Following treatment there was a reduction in SOL for all children except Matt. Parental presence during sleep onset was also eliminated during intervention for Matt, Catherine, Andrew and George. In the case of Harry, the parents were not continually present during the sleep onset period during any phase of the study.

Matt. For Matt, his SOL was consistently low during baseline (up to 5 minutes). After an initial small increase in SOL at the beginning of intervention (nights 10 and 11), Matt showed a consistent SOL until Day 41 when his mother left the room and his SOL escalated to 180 minutes. This largely resolved as the intervention continued, until P4 when he was moved to a new bed and it lengthened markedly again. His parents removed him from the study shortly after that.

Catherine. For Catherine, her SOL was variable during baseline (between 5 and 30 minutes). After an initial escalation of her SOL to 75 minutes during P1, this decreased to a SOL of 5 minutes or below with the introduction of P2 (milk removed during the bedtime routine), which lasted 8 nights. Her SOL then became variable again, with a peak of 60 minutes observed on Day 64 when her mother moved to the door. This largely resolved as the intervention continued, until Day 90 when her mothers' presence was removed and her SOL increased to 45 minutes. Her SOL then decreased to below baseline levels. Catherine's intervention was still continuing at the completion of the current study.

Andrew. Andrew's SOL during baseline was variable (between 5 and 80 minutes). He was given melatonin on the last night of baseline, following a long SOL period. His SOL remained variable but reasonable low (between 2 and 20 minutes) during P1, until his mother moved to the door, when his SOL increased to 40 minutes. Another increase in his SOL was seen when his mother moved to the hallway, with his SOL escalating to 90 minutes. This largely resolved during P2 as his bedtime was moved later, and remained consistent as his mothers' presence was removed. Andrew was provided with melatonin on 13 nights during P2, but this did not appear to effect his SOL. The variability in his SOL increased again during P3 when he was given a new bed. During the 7-week follow-up, Andrew's SOL varied between 10 and 20 minutes. He was provide with melatonin on all of these nights, and his mother sat by his bed for three of these nights.

George. For George, his SOL was variable during baseline (between 10 and 30 minutes), and remained variable during the first three phases of intervention, peaking to a SOL of 70 minutes following a period of illness at the end of P3. This largely resolved at P4, when his mothers' presence was completely removed and his Vallergran increased, and for the remainder of the duration of the intervention, with the exception of Day 81 when his Vallergran dosage decreased and his SOL was 30 minutes. His SOL was 5 minutes or below on 33/40 of these nights, which was a significant improvement on baseline. This improvement was maintained during the 6-week follow-up.

Harry. The reduction in SOL was most marked for Harry. During baseline, his SOL was the longest of all participants, lasting between 25 and 120 minutes. Immediately upon the introduction of intervention, his SOL reduced significantly. An increase in SOL was observed on Days 50 and 51, nights in which his

grandparents were babysitting and his bedtime was faded earlier. Thereafter, his SOL was greatly reduced, with Harry achieving a SOL of 7 minutes or less on 18/19 of the remaining nights. This was a significant improvement on baseline SOL durations, that was maintained during the 7 week follow-up.

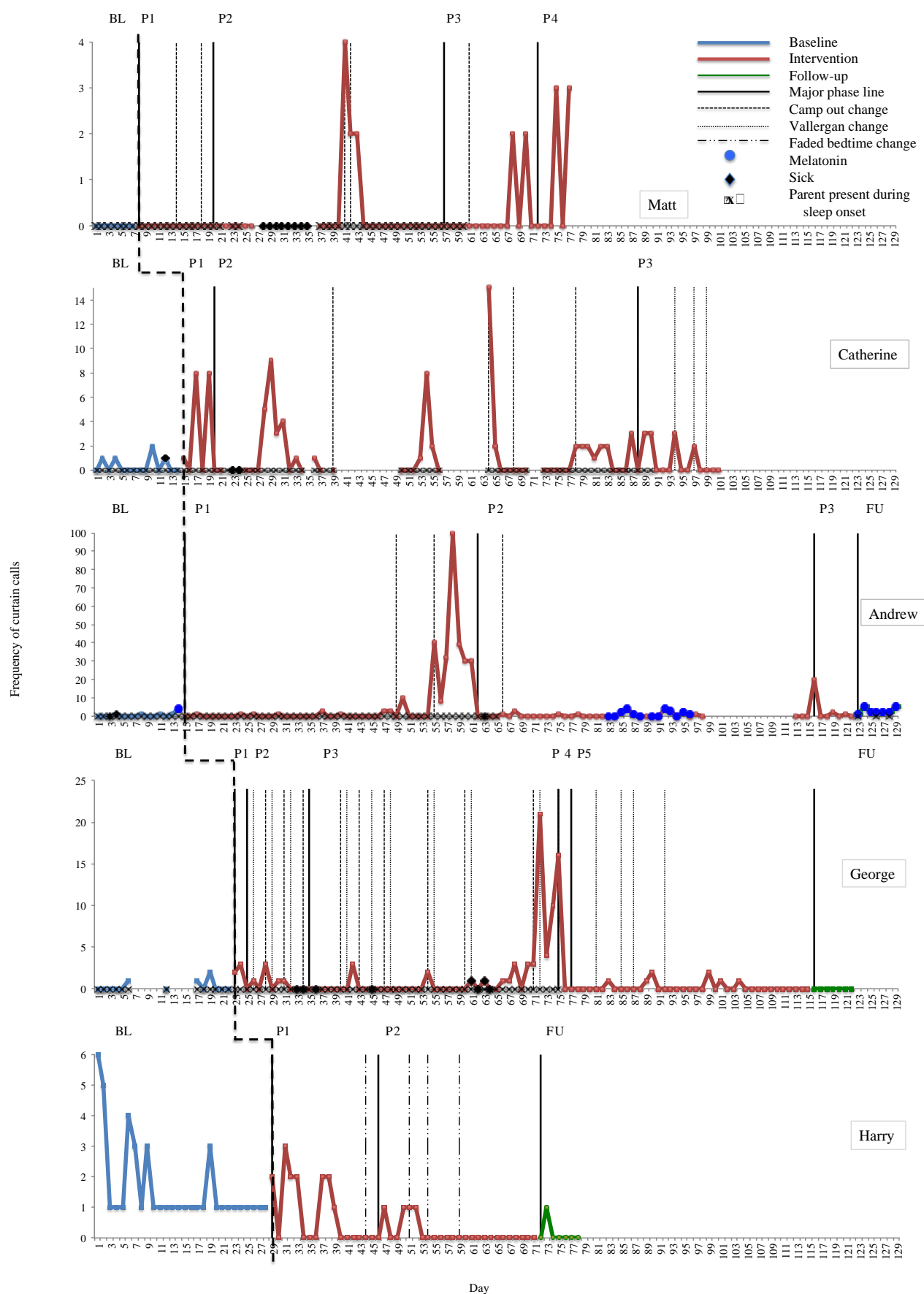


Figure 5. Frequency of curtain calls during baseline, intervention and follow-up for Study 2 participants.

Effect on curtain calls. Figure 5 presents baseline, intervention and follow-up data on the frequency of curtain calls (CC's) for each of the five children in the study. During baseline, the frequency of CC's was highly variable for Harry, somewhat variable for Catherine, Andrew and George, and consistently non-existent for Matt. Following intervention, there was a reduction in CC's for Harry, George and Catherine, while Andrew returned to baseline levels. Matt's frequency of CC's increased throughout his intervention.

Matt. Matt had no CC's during baseline, and he continued to have none until he had four on Day 41 when his mother left the room but was still visible. He had two CC's on the next two nights as his mother moved so only her legs were visible in the doorway, and then his frequency of CC's returned to zero. This continued until the last 10 days of intervention, when his frequency of CC's became more variable, following which his parents removed him from the study.

Catherine. For Catherine, her frequency of CC's varied between one and two during baseline. This escalated to up to eight CC's during P1, but decreased to zero with the introduction of P2 (milk removed from the bedtime routine), which lasted eight nights. Her frequency of CC's became variable again, with a peak of 14 CC's on Day 64 when her mother moved to the door. This largely resolved as the intervention continued, with peaks observed as her mother moved in to the hallway (Day 64) and when her Vallergan dose was decreased on Days 94 and 97. She had no CC's on 9/11 of the final nights of this study, which was an improvement on her baseline levels.

Andrew. During baseline, Andrew's frequency of curtain calls ranged between zero and four, with no CC's on 9/14 nights. This remained consistent until an increase to 10 CC's on Day 50 following his mother moving to the door. This

resolved until his mother moved to the hallway on Day 55, peaking at 100 CC's on Day 58. This largely resolved during P2 as his bedtime was moved later, and remained consistent as his mothers presence was removed, until P3, when there was a sudden increase of 20 CC's on the night that he got a new bed. This resolved again, to baseline levels for the remained of the intervention. During the 7-week follow-up, Andrew's frequency of CC's ranged between 2 and 5. He was provided with melatonin on all of these nights.

George. During baseline, George had between zero and two CC's, with none on 10/13 nights. This remained fairly consistent until Day 72, when he had 22 CC's as his mother became harder to see and his Vallergran decreased to 0mls. This remained elevated until Day 76, when his mothers' presence was removed completely. Thereafter, his frequency of CC's largely resolved, with no CC's observed on 34/40 nights, which was an improvement on baseline levels. During the 6-week follow-up, George had no CC's.

Harry. The reduction in CC's was most marked for Harry. During baseline, his frequency of CC's was between one and six. Between Days 29 and 39, his frequency of CC's remained variable. Thereafter, he had no CC's for the remainder of the intervention, with the exceptions of Days 47, and 50 to 52, where he had one CC on each occasion. On two of these nights, his grandparents babysat him. This was maintained at the 7-week follow-up, with the exception of Day 73 when he had one night with one CC.

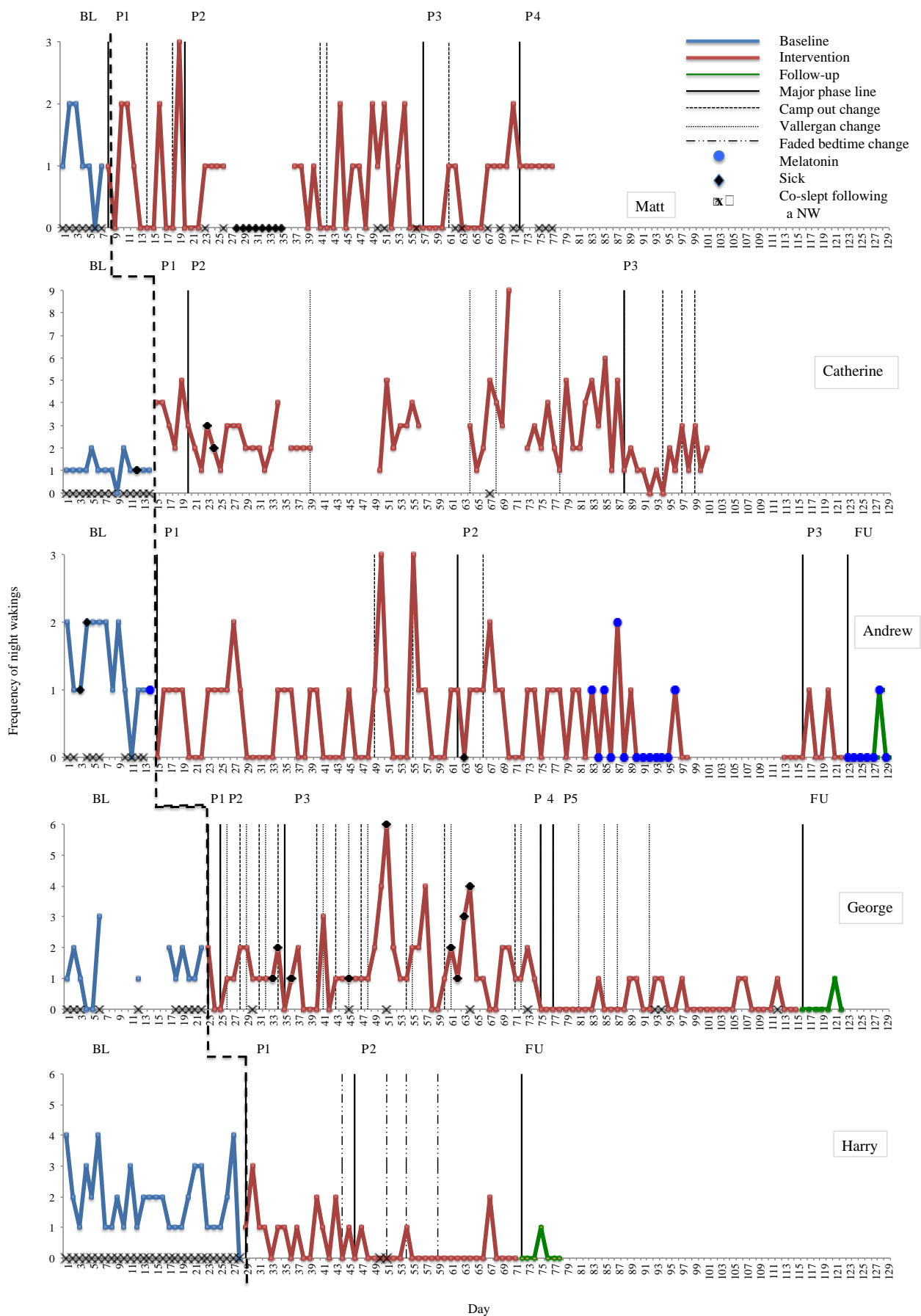


Figure 6. Frequency of night wakings during baseline, intervention and follow-up for Study 2 participants.

Effect on frequency of night wakings. Figure 6 presents baseline, intervention and follow-up data for frequency of night wakings (NW's) for each of the five children in the study. During baseline, the frequency of NW's was variable for all participants. Following treatment, there was a reduction in the frequency of NW's for all children except Catherine and Matt. Co-sleeping was also eliminated during intervention for Catherine, Andrew, George and Harry.

Matt. During baseline, Matt had between zero and two NW's. These NW's were always followed with co-sleeping. His frequency of NW's remained variable during intervention. He had a period of improvement from Days 55 to 66, where he only had two nights (Days 61 and 62) where a NW occurred, that followed the removal of his mothers presence. After Day 67, his frequency of NW's increased again, with 7/11 of these NW's being followed by co-sleeping. His parents removed him for the study shortly after that.

Catherine. Catherine had between zero and two NW's during baseline. Immediately upon the introduction of intervention, her NW's became more frequent and variable, peaking at nine NW's on Day 70. A slight decrease was observed during P3 (introduction of Vallergran), with Catherine having no NW's on two nights. The highest number of NW's during P3 was for Days 97 and 99, which coincided with decreases in Vallergran dosage. At the completion of the study, Catherine's frequency of NW's was nearing a return to baseline levels.

Andrew. For Andrew, his frequency of NW's during baseline ranged between zero and two, with no NW's on just 1/14 nights. During P1 of intervention, his frequency of NW's improved, with no NW's on 24/41 nights. However, increases to three NW's were seen on days 50 and 55, when his mother moved to the door, and then in to the hallway. His frequency of NW's remained

variable for the remainder of his intervention, but there were periods of no NW's from Day 90-95 (when he had melatonin), and for four days following his return from holiday (Days 113-116). During his 7-week follow-up, he had a NW on just 1/7 nights. He was provided with melatonin on all of these nights.

George. During baseline, George had between zero and three NW's, with no NW's on just 2/13 nights. This remained variable during his intervention, until P4 when he could no longer see his mother and his Vallergan was increased. For the remainder of the intervention, George's frequency of NW's largely resolved, with him having no NW's on 32/41 nights. However, three of the nights when he had NW's were followed by co-sleeping. This was a significant improvement compared to baseline, which was maintained at the 6-week follow-up.

Harry. For Harry, his frequency of NW's during baseline was between zero and four. During Days 29 to 48, his frequency of NW's remained variable. Thereafter, he had no NW's for the remainder of the intervention, with the exceptions of one on Day 54, when his bedtime was faded back, and two on Day 67, when he had had a day nap. This was a significant reduction from baseline. Improvements were maintained at follow-up, with only one NW on Day 75.

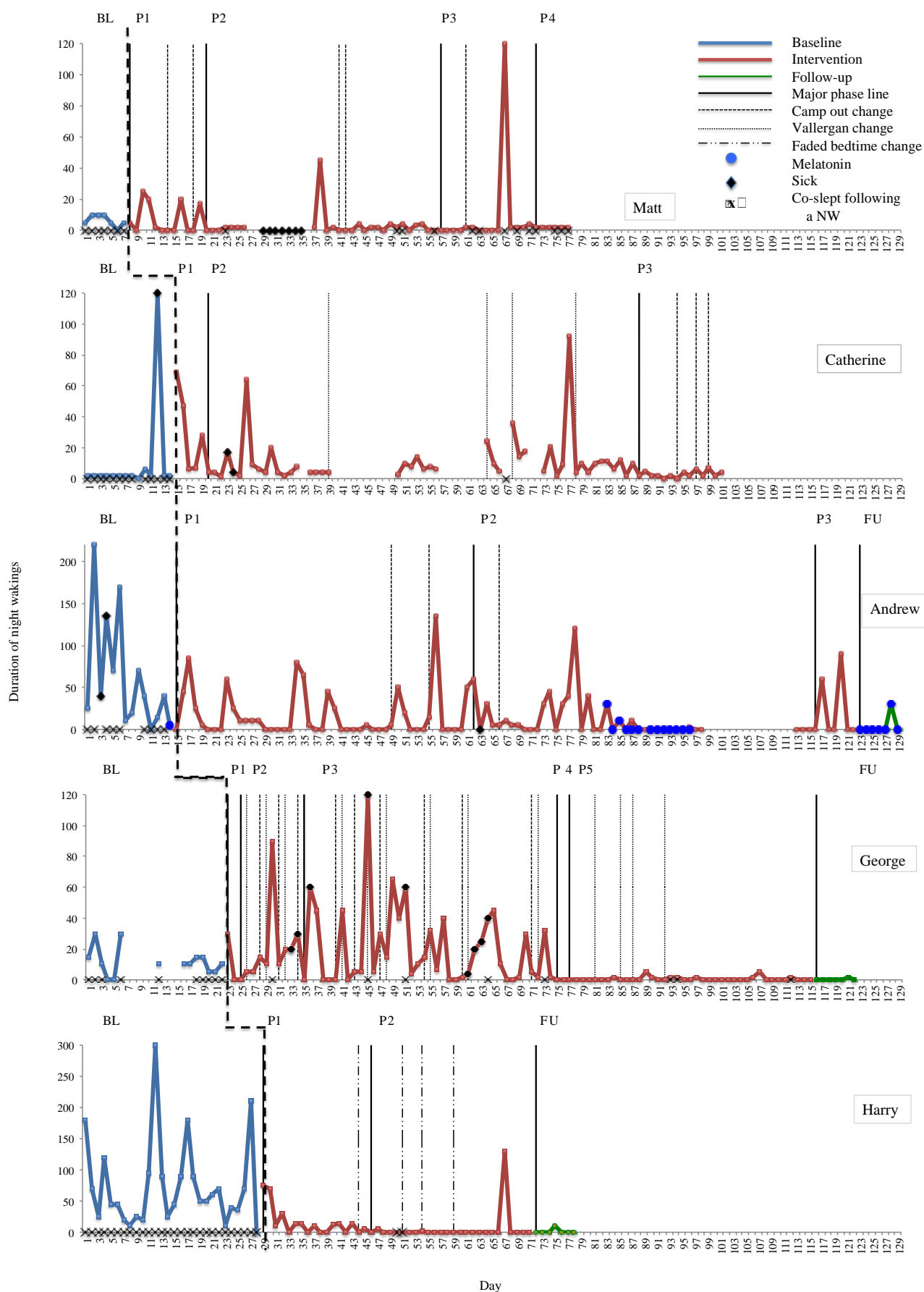


Figure 7. Duration of night wakings during baseline, intervention and follow-up for Study 2 participants.

Effect on duration of night wakings. Figure 7 represents baseline, intervention and follow-up data for duration of NW's for each of the five children included in this study. During baseline, duration of NW's was highly variable for Andrew and Harry, and less variable for Catherine, Matt and George. Following treatment, there was a reduction in the duration of NW's for all children, except Catherine, who had not yet finished data collection.

Matt. During baseline, Matt's duration of NW's varied between 0 and 45 minutes, and were all followed by co-sleeping. After an initial small increase in the duration of NW's at the beginning of intervention (durations between 17 and 25 minutes on Days 10, 11, 16 and 18), Matt's duration of NW's largely resolved for the remainder of the intervention, with the exceptions of Days 38 (when he woke for 45 minutes) and 67 (when he woke for 120 minutes).

Catherine. For Catherine, her duration of NW's during baseline were very short (typically 2 minutes) with the exception of Day 12 where she was awake for 120 minutes. She was sick on this night however. Co-sleeping followed all night wakings during baseline. Her duration of NW's was variable during intervention (between 0 and 90 minutes), but became shorter and less variable after night 78, when her mother moved out to the hallway (between 0 and 11 minutes). At the end of this study, Catherine's duration of NW's had returned to near baseline levels.

Andrew. Andrew's baseline duration of NW's varied between 0 and 220 minutes, and were frequently followed by co-sleeping. Throughout his intervention, Andrew had an increase in the number of nights where he did not wake. On the nights that he did wake, his duration of NW's was mostly below baseline levels. This was maintained at the 7-week follow-up.

George. During baseline, George's duration of NW's ranged between 0 and 30 minutes, and typically resulted in co-sleeping. Duration of NW's was initially variable following the introduction of interventions, until P4 when his mother's presence was moved and his Vallerger increased. For the remainder of his intervention, on the nights that he did wake, his duration of NW's did not exceed 5 minutes. This is a marked improvement compared to baseline. During the 6-week follow-up, his one NW lasted 2 minutes, during which he was being comforted following a distressing nightmare.

Harry. For Harry, his duration of NW's during baseline were variable, lasting between 0 and 300 minutes and always resulting in co-sleeping. Although his duration of NW's was still variable until Day 47, the amount of time he was awake for was significantly lower (between 0 and 75 minutes). From Day 48 until intervention concluded, Harry had no NW's, with the exception of Days 54 and 67, which he woke for 2 minutes and 130 minutes respectively. This is a significant improvement from baseline, which was maintained at follow-up.

IOA data. IOA data was obtained for 22%, 22% and 25% of Andrew's, George's and Harry's days respectively, with the percentage of agreement being 94%, 81% and 72 % respectively.

Frequency of returns to bed for Andrew.

Figure 8 presents baseline, intervention and follow-up data for the number of times Andrew's mother returned Andrew to his bed during sleep onset, during the night, and in the morning. This represents the intensity of his responses to the changes implemented through intervention.

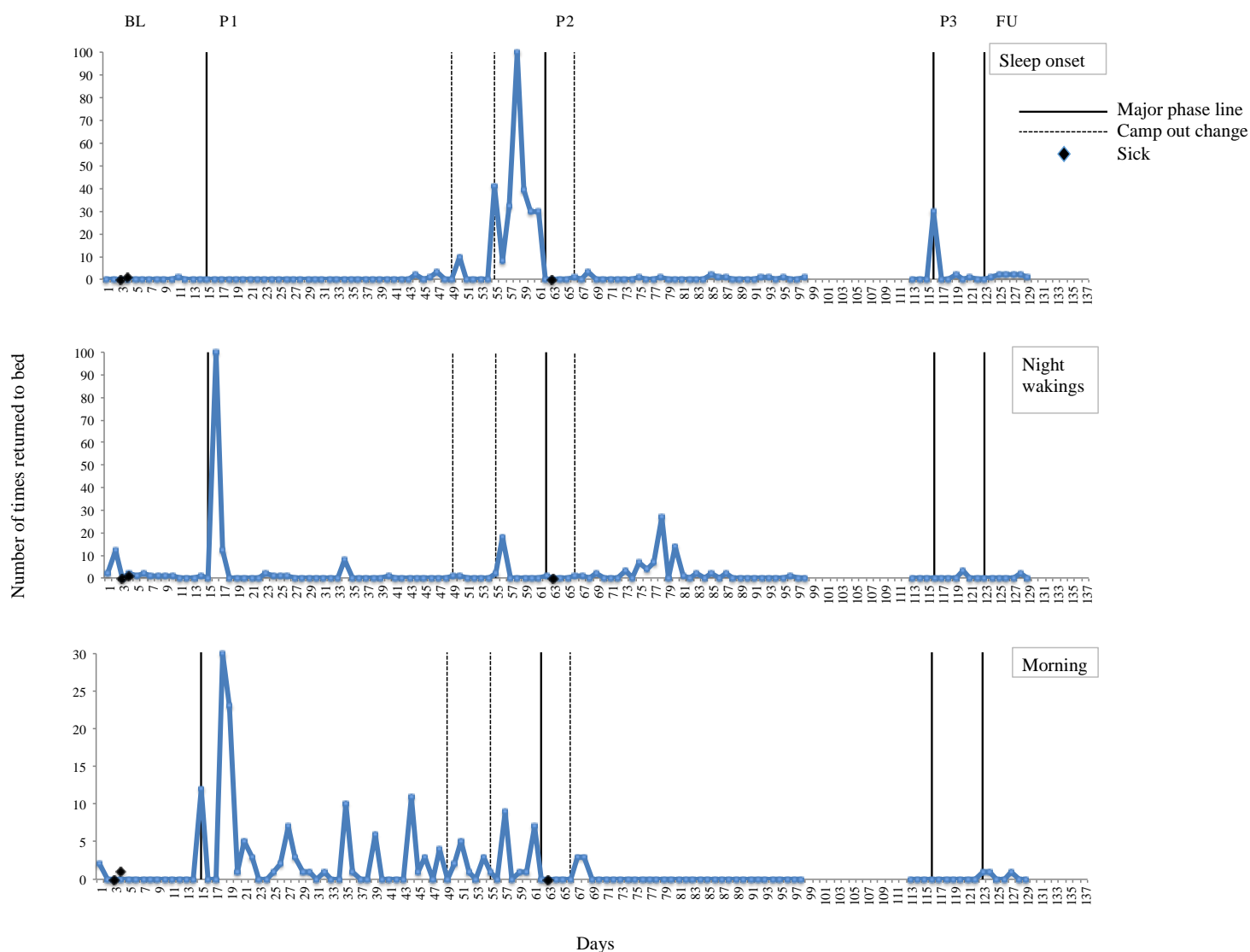


Figure 8. The number of times Andrew was returned to his bed at sleep onset, during the night, and in the morning, during baseline, intervention, and follow-up.

Sleep onset. At sleep onset, Andrew was returned to bed just once on one night during baseline. On this night, he was not at home, and left his room to seek out his mother. During intervention, Andrew did not need to be returned to bed until Day 44, when his number of returns to bed became variable. An increase in returns to bed was observed after his mother moved to the door. This escalated after she moved to the hallway, peaking at 100 returns. This largely resolved after P2, when his bedtime was moved back, and remained consistent as his mothers presence was removed, until night 116 when he was moved to a new bed. On this

night, he was returned to bed 30 times, but this did appear to settle again for the remainder of the intervention. During follow-up, Andrew was returned to bed between 0 and 2 times per night.

Night wakings. During the night, the number of times Andrew was returned to bed during baseline was consistently low, with the exception of Day 2, when he was returned to bed 12 times. After an initial sharp escalation to 100 times returned to bed on the second night of intervention, this largely returned to baseline levels, until day 56 when he was returned to bed 20 times after his mother moved to the hallway. The frequency with which he was returned to bed during the night became more variable during Days 73 to 87, peaking at 30 times returned to bed, but then resolved again to baseline levels for the remainder of the intervention, and continued through follow-up.

Morning. Andrew was not returned to bed in the morning during baseline, with the exception of night one when he was returned two times. Immediately upon the introduction of intervention, the number of times he was returned to bed increased significantly, peaking at 30 times on Day 18. This remained variable until Day 62 when his bedtime was faded back. With the exceptions of Days 67 and 68, Andrew was not returned to bed in the morning for the remainder of the intervention. At follow up, he was returned to bed on three nights, and zero times on the remaining four nights.

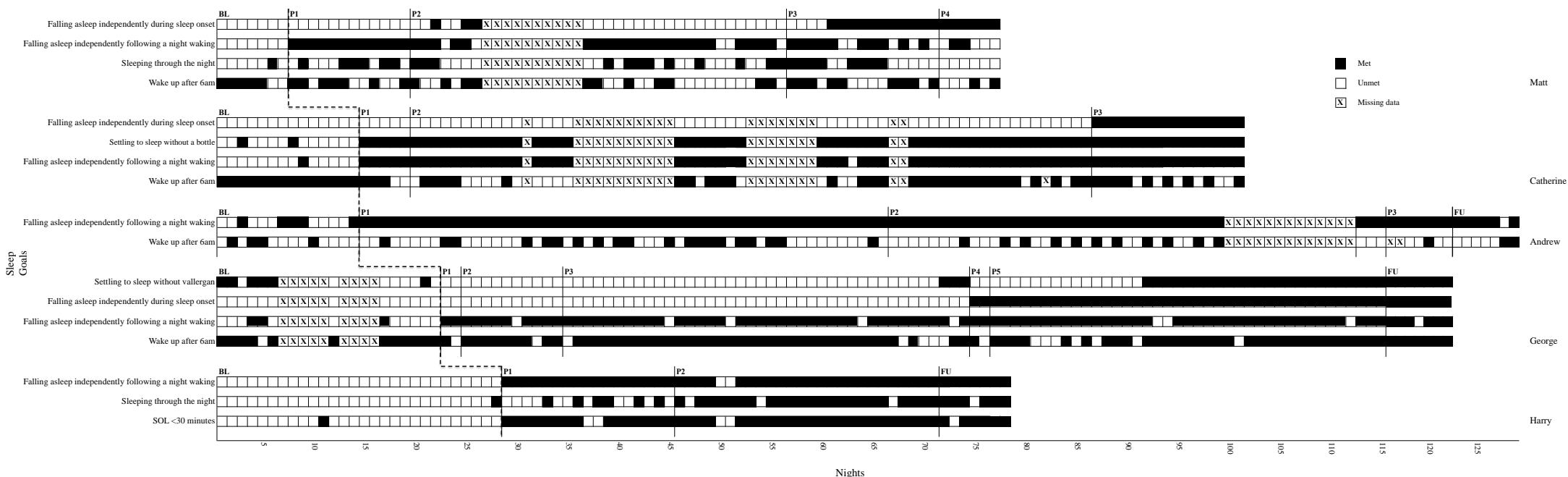


Figure 9. Goal attainment across baseline, intervention and follow-up for Study 2 participants.

Goal attainment. Figure 9 depicts whether sleep goals were met across individualised sleep-related measures for each child during baseline, intervention and follow-up. The goal chart conveys the extent to which sleep goals were achieved, and helps to convey whether the child was a more independent sleeper following treatment. All families had the goal to not co-sleep following a night waking. No parental presence during sleep onset was a shared goal for Catherine, Matt and George, and waking up after 6am was a goal shared by Catherine, Matt, Andrew, and George. Most children had additional measures reflecting their personal goals. Matt's was to have no night wakings, Catherine's was to not use a bottle to settle, George's was to have no Vallergan, and Harry's additional measures were to have no night wakings and a sleep onset latency of less than 30 minutes. The criteria for meeting the sleep goals were explained under each individual's methods in the study 2 methods section of this thesis.

For all five children, there were more nights in which sleep goals were met at the end of their treatment than in baseline (percentages of sleep goals met during baseline nights were 21%, 36%, 38%, 41% and 2% for Matt, Catherine, Andrew, George and Harry respectively; percentages of sleep goals met for the last ten treatment nights were 50%, 88%, 39%, 98%, and 97% for Matt, Catherine, Andrew, George and Harry respectively), demonstrating a positive effect on multiple sleep outcome measures as a function of individualised interventions.

Matt. For Matt, during baseline and phase one, he never achieved his goal of falling asleep independently during sleep onset. Due to the camping out procedure, this goal was not consistently achieved until Day 61. Until then, Matt had three nights where he did not have his parents' presence during sleep onset. On two of these nights, he fell asleep on the couch, and the third he fell asleep while his

parents attended to their infant. His second goal of falling asleep independently following a night waking was not achieved during baseline. During Phase one, he did not co-sleep. However, after this, Matt showed a progressive reduction in obtaining this goal throughout the remainder of his treatment duration. However, it was an improvement on baseline goal attainment. He achieved his goal of sleeping through the night once during baseline. Significant improvements were seen during Phases one to three, with Matt not waking on 26/60 nights (43%). However, he had night wakings consistently for the last 11 nights. For his fourth goal of waking up after 6am, he achieved this on 5/7 nights (71%) during baseline, which decreased to 29/60 nights (48%) during intervention.

Catherine. For Catherine, the goal of falling asleep independently during sleep onset was not met during baseline. Due to the camping out procedure, this goal was not achieved until day 87, but was consistently achieved thereafter. Catherine required her bottle to settle on 12/15 nights during baseline. This was eliminated with the introduction of intervention on Day 15, and she consistently achieved this goal for the remainder of the intervention. Catherine consistently achieved her goal of waking after 6am during baseline. This decreased to 41/46 nights (62%) during intervention.

Andrew. During baseline, Andrew achieved his goal of falling asleep independently following a night waking on 5/15 nights. During intervention, he consistently achieved this goal, and eliminated co-sleeping. His goal of waking after 6am was more variable during baseline, and remained variable throughout intervention. Post-intervention, Andrew required parental presence to fall asleep on one night following a night waking. His goal of waking after 6am remained variable.

George. For George, his goal of settling to sleep without Vallergan was achieved on 6/13 of recorded nights (46%) during baseline. Due to the Vallergan regimen, this was not consistently achieved during intervention until Day 92. He did not achieve his goal of falling asleep independently at sleep onset during baseline, and due to the camping out procedure, this goal was not achieved until day 75, but was consistently achieved thereafter. For his goal of falling to sleep independently following a night waking, George achieved this on 3/13 of recorded nights (23%) during baseline. Two of these nights he had no night wakings and the third he has being babysat. During intervention, he achieved this goal on 85/93 of nights (91%). Two of the nights when he co-slept followed two long night wakings, two occurred on days he was sick, two nights his mother found him sleeping with his sister in her bed, one occurred after multiple night wakings, and one was initiated by his mother due to her concerns about his breathing following a swimming accident. The goal of him not waking after 6am was achieved on 12/13 of nights during baseline and was achieved on 78/93 mornings during intervention. From Day 92, he consistently woke after 6am, with the exception of Day 101, which was Christmas morning. At the 6-week follow-up, George continued to meet his goals of falling asleep without Vallergan, falling asleep independently during sleep onset, and waking after 6am. On one night, he required his mothers presence to resettle following a night waking.

Harry. For Harry, his goals of settling to sleep independently after a night waking, sleeping through the night, and a SOL of less than 30 minutes were not met during baseline, with the exception of Day 11 when he fell asleep in 25 minutes, and Day 28 in which he had no night wakings. Significant improvements in these goals were observed during intervention. He did not co-sleep, with the exception of Days 50 and 51, when his grandparents were babysitting him and they let him

sleep with them. From Day 48, he had no night wakings, with the exception of Days 54 and 67. Harry fell asleep within 30 minutes on 38/42 of nights (90%). For two of the four nights which he did not achieve this goal, he was asleep within 32 and 35 minutes. On the other two nights, his grandparents were babysitting. Post intervention, Harry did not co-sleep, but he did have one night waking on one night. He achieved his goal of a SOL of less than 30 minutes every night, with the exception of Day 73 when it took him 35 minutes to get to sleep.

Table 5. Comparison of Pre- and Post-Intervention Scores on the CSHQ for Study 2 Participants

Variable scores	Matt		Catherine		Andrew		George		Harry	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Bedtime Resistance	12	N/A	11	N/A	7	9	12	7	10	7
Sleep Onset Delay	1	N/A	1	N/A	1	1	1	1	3	3
Sleep Duration	3	N/A	3	N/A	7	7	7	3	7	4
Sleep Anxiety	8	N/A	8	N/A	6	7	9	6	8	4
Night Wakings	9	N/A	8	N/A	4	4	7	6	8	6
Parasomnias	9	N/A	12	N/A	7	7	13	16	8	9
Disordered Breathing	3	N/A	3	N/A	3	3	4	3	3	3
Daytime Sleepiness	11	N/A	10	N/A	8	9	7	7	10	10
Total Difficulties	50	N/A	51	N/A	41	43	54	47	54	42

(Note: bold = above average scores)

Child Sleep Habits Questionnaire. Results of the CSHQ (Owens et al., 2000) are presented in Table 5. A higher score indicates more difficulties in that area. These results show that before intervention, Matt, George, Harry and Catherine were all rated by their parents as having above average total sleep difficulties. Following intervention, parents total sleep difficulty ratings reduced and were within average levels for George and Harry, but Andrew's parents reported a slight increase in total sleep difficulties.

Matt and Catherine. Pre-treatment scores for Matt and Catherine indicated that they both had above average total sleep difficulties (50 and 51 respectively), as well as above average bedtime resistance (Matt: 12, Catherine: 11), sleep anxiety (both 8), and night waking (Matt: 9, Catherine: 8) difficulties. Catherine also scored above average for parasomnias (12). All other scores were within the normal range. Post-treatment scores on the CSHQ were not collected for Matt and Catherine.

Andrew. For Andrew, his bedtime resistance and sleep anxiety scores increased from the average to above average ranges following intervention (from 7 to 9 and 6 to 7 respectively). His daytime sleepiness score also increased (from 8 to 9), but remained in the average range. All other variables remained constant. Overall his total sleep difficulties scores increased from 41 to 43, but stayed within the average range.

George. For George, these results showed reductions from above average to average scores for bedtime resistance, sleep duration, and sleep anxiety (from 12 to 7, 7 to 3, and 9 to 6 respectively). Reductions were also observed but remained above average for night wakings (from 7 to 6). A decrease was seen for disordered breathing (from 4 to 3). Scores remained consistent for sleep onset delay (1), and

daytime sleepiness (7), and parasomnias increased from 13 to 16. Overall, his total sleep difficulties score reduced from an above average score of 54 to an average score of 47.

Harry. For Harry, these results showed reductions from above average to average scores for bedtime resistance (from 10 to 7), sleep duration (from 7 to 4), and sleep anxiety (from 8 to 4). Pre-intervention, his night waking's were above average, which reduced post-intervention, but remained above average (from 8 to 6). Scores remained consistent for sleep onset delay (3), disordered breathing (3), and daytime sleepiness (10), but parasomnias increased (from 8 to 9). Overall, his total sleep difficulties score reduced from an above average score of 54 to an average score of 42.

Parent Wellbeing Measures

DASS-21. The results of the DASS-21 for the parents of all Study 2 participants are presented in Table 6. A higher score indicates a greater likelihood of psychological distress in that area. These results indicate that depression scores either decreased or remained constant following intervention for all parents of children who completed intervention, but anxiety and stress scores were variable. The most marked improvements were for George's mother.

Matt and Catherine. Both Matt and Catherine's parents scored in the normal range across the depression, anxiety and stress dimensions pre-intervention. Post-treatment scores on the DASS-21 were not collected for Matt and Catherine.

Andrew. Andrew's parents both scored in the normal range for all dimensions pre- and post-intervention. Both of his parent's depression scores decreased slightly

(from 3 to 2 and 5 to 4 for his mother and father respectively). His mother's anxiety score remained constant at 1, while his father's score increased from 1 to 2. His mothers stress score decreased from 8 to 5, while his fathers increased form 4 to 6.

George. George's mother scored in the moderate range for depression pre-intervention. This decreased to the normal range post-intervention, with a decrease in scores from 17 to 7. She scored in the mild range for both anxiety and stress pre-intervention. Following intervention, her anxiety score decreased to the normal range, with a score of 8 to a score of 4, and her stress score decreased to the normal range, with a score of 17 to a score of 10. No data was collected for George's father, as he is not involved in George's everyday life.

Harry. Harry's parents scored in the normal range for depression, anxiety and stress pre-intervention. Following intervention, his father's scores all remained within the normal range, with slight decreases seen in his depression score (3 down to 2) and his anxiety score (2 down to 1). However, a slight increase was seen in his stress score (5 up to 8). Harry's mother's depression score remained constant, with a score of 2 both pre- and post-intervention. Her anxiety score increased to the mild range post-intervention, with a score increase of 5 to 8. Her stress score also increased post-intervention, from 3 to 4, but remained within the normal range.

Table 6. Comparison of Pre-and Post-Intervention Scores on the DASS-21 for Study 2 Participants

Variable scores	Matt				Catherine				Andrew				George				Harry			
	Mother		Father		Mother		Father		Mother		Father		Mother		Father		Mother		Father	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Depression	2	N/A	2	N/A	0	N/A	0	N/A	3	2	5	4	17	7	N/A	N/A	2	2	3	2
	(N)		(N)		(N)		(N)		(N)	(N)	(N)	(N)	(MO)	(N)			(N)	(N)	(N)	(N)
Anxiety	0	N/A	4	N/A	0	N/A	0	N/A	1	1	1	2	8	4	N/A	N/A	5	8	2	1
	(N)		(N)		(N)		(N)		(N)	(N)	(N)	(N)	(MI)	(N)			(N)	(MI)	(N)	(N)
Stress	1	N/A	7	N/A	5	N/A	4	N/A	8	5	4	6	17	10	N/A	N/A	3	4	5	8
	(N)		(N)		(N)		(N)		(N)	(N)	(N)	(N)	(MI)	(N)			(N)	(N)	(N)	(N)

(Note: N=Normal range; MI=Mild range; MO=Moderate range)

Table 7. Comparison of Pre-and Post-Intervention Scores on the PSQI for the Parents of Study 2 Participants

	Matt				Catherine				Andrew				George				Harry				
	Mother		Father		Mother		Father		Mother		Father		Mother		Father		Mother		Father		
Variable scores	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Max
Subjective sleep quality	3	N/A	2	N/A	2	N/A	1	N/A	2	1	2	1	3	1	N/A	N/A	2	1	2	1	3
Sleep latency	0	N/A	1	N/A	2	N/A	2	N/A	2	1	2	2	3	3	N/A	N/A	1	1	3	2	3
Sleep duration	1	N/A	0	N/A	1	N/A	0	N/A	1	1	1	0	3	1	N/A	N/A	1	0	1	0	3
Habitual sleep efficiency	0	N/A	0	N/A	0	N/A	0	N/A	3	0	1	0	3	2	N/A	N/A	2	0	1	0	3
Sleep disturbances	1	N/A	2	N/A	2	N/A	1	N/A	1	1	2	1	1	2	N/A	N/A	1	1	2	2	3
Use of sleeping medication	0	N/A	0	N/A	0	N/A	0	N/A	0	0	1	1	0	1	N/A	N/A	0	0	0	0	3
Daytime dysfunction	0	N/A	2	N/A	0	N/A	2	N/A	1	0	1	1	3	2	N/A	N/A	1	1	1	2	3
Global PSQI score	5	N/A	7	N/A	7	N/A	6	N/A	10	4	9	6	16	12	N/A	N/A	8	4	10	7	21

PSQI. The results of the PSQI for the parents of all study 2 children are presented in Table 7. This gives an indication of the parent's sleep quality pre- and post-intervention. A lower score indicates better sleep quality. The parents that completed the intervention showed an improvement in their global sleep quality between the two time points.

Matt. Matt's mother and father had global sleep quality scores of 5/21 and 7/21 at pre-intervention respectively. Pre-intervention, Matt's mother rated her sleep quality to be very bad (3/3) and his father rated his to be fairly bad (2/3). Sleep latency, sleep duration, and habitual sleep efficiency was rated by both parents as very good or fairly good. Neither parent used any sleeping medications. Matt's mother rated her sleep disturbances as fairly good (1/3) whereas his father rated his sleep disturbances as fairly bad (2/3), and his mother rated her daytime dysfunction as very good (0/3), but his father rated his daytime dysfunction as fairly bad (2/3). Post-treatment scores on the PSQI were not collected for Matt's parents as intervention was not completed.

Catherine. Before intervention, Catherine's mother and father had global sleep quality scores of 7/21 and 6/21 respectively. Pre-intervention, Catherine's mother and father did not rate any of the dimensions as very bad. They both rated habitual sleep efficiency and use of sleeping medication as very good (0/3), and sleep latency as fairly bad (2/3). Catherine's mother rated both her subjective sleep quality and sleep disturbances as fairly bad (2/3), and her father rated both of these dimensions as fairly good (1/3). Catherine's mother and father rated their sleep duration as fairly good (2/3) and very good (0/3) respectively, and daytime dysfunction was rated as very good (0/3) and fairly bad (2/3) for her mother and father respectively.

Post-intervention scores on the PSQI were not collected for Catherine's parents, as intervention had not yet been completed.

Andrew. For Andrew's parents, both his mothers and fathers global sleep quality scores decreased between pre- and post-intervention, from 10/21 to 4/21 and 9/21 to 6/21 respectively. Decreases between pre- and post-intervention were also seen on the subjective sleep quality domain (both parents: 2/3 down to 1/3), and habitual sleep efficiency domain (mother: 3/3 down to 0/3, father: 1/3 down to 0/3) for both parents. Scores for both parents remained constant on the use of sleeping medications domain (mother: 0/3, father 1/3). Scores for Andrew's mother also decreased on the sleep latency (from 2/3 to 1/3) and daytime dysfunction domains (from 1/3 to 0/3), while his fathers scores on these domains remained constant (2/3 and 1/1 respectively), whereas scores for Andrew's father decreased on the sleep duration (from 1/3 to 0/3) and sleep disturbances (from 2/3 to 1/3) domains, while his mothers scores on these domains remained constant (1/3 for both domains). There were no domains where scores increased for either parent.

George. For George's mother, her global sleep quality score decreased from 16/21 to 12/21 between pre- and post-intervention. Decreases were seen on the subjective sleep quality domain (from 3/3 to 1/3), sleep duration domain (from 3/3 to 1/3, habitual sleep efficiency domain (from 3/3/ to 2/3), and daytime dysfunction domain (from 3/3 to 2/3). Scores remained consistent for George's mother on the sleep latency domain (3/3). His mother's scores reflect in increase in sleep disturbances (from 1/3 to 2/3), and an increase in use of sleep medication (from 0/3 to 1/3).

Harry. For Harry's parents, both his mother and father's global sleep quality scores decreased between pre- and post-intervention, from 8/21 to 4/21 and 10/21

to 7/21 for his mother and father respectively, indicating an improvement in overall sleep. Decreases between pre- and post-intervention were seen on the subjective sleep quality (both parents: 2/3 down to 1/3), sleep duration (both parents: 2/3 down to 1/3), and habitual sleep efficiency (mother: 2/3 down to 0/3, father: 1/3 down to 0/3) domains for both parents. Scores for both parents remained consistent from pre- to post- intervention on the sleep disturbances domain (mother: 1/3, father: 2/3), and neither parent used sleeping medications at either time point. Harry’s mothers sleep latency and daytime dysfunction scores did not change between pre- and post-intervention, all remaining fairly good (1/3). Harry’s father reported a decrease in his sleep latency problems, from very bad to fairly bad (3/3/ to 2/3), but his daytime dysfunction increased from fairly good to fairly bad (1/3 to 2/3).

RQI. Table 8 presents the results of the RQI at pre- and post-intervention for the parents of all Study 2 children except George, as his mother was not in a relationship. A higher score indicates greater satisfaction. The global RQI score indicates the level of partner satisfaction for each individual. Results are mixed.

Table 8. Comparison of Pre-and Post-Intervention Scores on the RQI for parents of Study 2 Participants

	Matt		Catherine		Andrew				Harry				Max
	Mother	Father	Mother	Father	Mother		Father		Mother		Father		
	Pre	Pre	Pre	Pre	Pre	Post	Pre	Post	Pre	Post	Pre	Post	
Global RQI	36	26	33	32	44	42	36	27	44	44	41	41	45

Matt. Matt's mother indicated a higher level of partner satisfaction than her husband (36/45 and 26/45 respectively). Post-intervention comparisons were not made, as Matt did not finish the intervention.

Catherine. Catherine's parents indicated similar levels of partner satisfaction (mother: 33/45, father: 32/45). Post-intervention comparisons were not made, as Catherine had not yet finished intervention.

Andrew. Andrew's mother indicated a very high level of partner satisfaction (44/45) pre-intervention, that decreased slightly (down to 42/45) post-intervention, indicating a slight reduction in satisfaction with her relationship. His father's pre-intervention score of 36/45 decreased to 27/45 post-intervention, indicating a reduction in his partner satisfaction.

Harry. Harry's parents both reported high levels of partner satisfaction (mother: 44/45, father: 41/45), which remained unchanged between pre- and post-intervention.

Child's Daytime Behaviour

GARS-3. The results of the GARS-3 at pre- and post-intervention for all Study 2 children are presented in Table 9. A higher score indicates a higher level of symptom severity. For all children who completed intervention, small reductions were seen in their overall ASD symptom severity scores, as reported by their parents.

Table 9. Comparison of Pre-and Post-Intervention Scores on the GARS-3 for Study 2 Participants

	Matt				Catherine				Andrew				George				Harry			
	Pre		Post		Pre		Post		Pre		Post		Pre		Post		Pre		Post	
Subscale performance	SS	PR	SS	PR	SS	PR	SS	PR	SS	PR	SS	PR	SS	PR	SS	PR	SS	PR	SS	PR
Restricted/Repetitive Behaviours	9	37	N/A	N/A	10	50	N/A	N/A	13	84	12	75	12	75	11	63	10	50	9	37
Social Interaction	12	75	N/A	N/A	11	63	N/A	N/A	12	75	9	37	9	37	7	16	12	75	12	75
Social Communication	11	63	N/A	N/A	11	63	N/A	N/A	12	75	12	75	12	75	12	75	12	75	11	63
Emotional Responses	7	16	N/A	N/A	11	63	N/A	N/A	11	63	10	50	12	75	13	84	12	75	11	63
Cognitive Style	7	16	N/A	N/A	9	37	N/A	N/A	7	16	7	16	8	25	11	63	8	25	10	50
Maladaptive Speech	6	9	N/A	N/A	8	25	N/A	N/A	9	37	10	50	15	95	12	75	11	63	11	63
Composite Score	89		N/A		100		N/A		106		100		112		109		108		106	

(Note: SS=scale score; PR=percentile rank)

Matt. Before intervention, Matt's parents gave him a composite score of 89, suggesting that it was 'very likely' he had ASD, and that he 'required substantial support'. His greatest difficulties appeared to be in the social interaction and social communication domains, where he scored higher than 75% and 63% of the population respectively. Post-intervention scores were not collected, as Matt did not complete intervention.

Catherine. Catherine's parent gave her a composite score of 100 before intervention, suggesting that that it was 'very likely' she had ASD, and that she 'required substantial support'. Her greatest difficulties appeared to be in the social interaction, social communication and emotional responses domains, where she scored higher than 63% of the population on each domain. Post-intervention scores have not yet been collected, as Catherine is still to complete intervention.

Andrew. Andrew had a pre-intervention composite score of 106, indicating that that it was 'very likely' he had ASD, and that he 'required very substantial support'. This decreased to a score of 100 following intervention, indicating a reduction in severity level to 'requiring substantial support'. Reductions were seen in his restricted/repetitive behaviours (PR of 84 down to 75), social interaction behaviours (PR of 75 down to 37), and emotional responses (PR of 63 down to 50), suggesting an improvement in these areas. However, his social communication scores and cognitive style remained consistent (PR's of 75 and 16 respectively), indicating no changes in these areas. However, his scores on the maladaptive speech subscale increased (PR of 37 up to 50), suggesting a decline in this area.

George. George had a pre-intervention composite score of 112, indicating that it was 'very likely' he had ASD, and that he 'required very substantial support'. This decreased to 109 following intervention, indicating a slight improvement in his

overall ASD symptomatology. Reductions were seen in his restricted/repetitive behaviours (PR of 75 down to 63), social interaction behaviours (PR of 37 down to 16) and maladaptive speech (PR of 95 down to 75), suggesting an improvement in these areas. No changes were reported in the area of social communication (PR of 75), however, increased were seen in his emotional responses (PR of 75 up to 84) and cognitive style (PR of 25 up to 63) behaviours, suggesting a decline in these areas.

Harry. Harry had a pre-intervention composite score of 108, indicating that it was 'very likely' he had ASD, and that he 'required very substantial support'. This decreased to 106 post-intervention, suggesting a slight improvement in his overall ASD symptomatology. Reductions from pre- to post-intervention were seen in his restricted/repetitive behaviours (PR of 50 down to 37), social communication (PR of 75 down to 63), and emotional responses (PR of 75 down to 63). His parents noted no changes in his social interaction and maladaptive speech performance from pre- to post-intervention (PR of 75 and 63 respectively), but increases were seen in his cognitive style (PR of 25 up to 50), suggesting some worsening in this area.

CBCL.

The results of the CBCL at pre- and post-intervention for Study 2 children are presented in Table 10. The CBCL identifies possible behavioural and emotional problems in children as rated by their parents. A higher score indicates more difficulties in that area.

Table 10. Comparison of Pre-and Post-Intervention T-scores on the CBCL for Study 2 Participants

	Matt				Andrew				George				Harry				Catherine			
	Pre		Post		Pre		Post		Pre		Post		Pre		Post		Pre		Post	
<i>Empirically Based Scales</i>	Score	R	Score	R	Score	R	Score	R	Score	R	Score	R	Score	R	Score	R	Score	R	Score	R
Emotionally Reactive	2	N	N/A		4	N	3	N	3	N	7	B	15	C	16	C	7	B	N/A	
Anxious/Depressed	5	N	N/A		6	N	3	N	3	N	7	B	10	C	11	C	6	N	N/A	
Somatic Complaints	0	N	N/A		4	N	2	N	2	N	7	C	6	B	8	C	5	B	N/A	
Withdrawn	8	C	N/A		7	C	6	C	6	C	4	N	7	C	9	C	10	C	N/A	
Sleep Problems	4	N	N/A		4	N	4	N	7	N	8	B	10	C	7	N	8	B	N/A	
Attention Problems	5	N	N/A		3	N	0	N	9	C	7	C	8	C	5	N	7	C	N/A	
Aggressive Behaviour	17	N	N/A		16	N	11	N	21	B	15	N	26	C	20	N	21	B	N/A	
Other Problems	14		N/A		17		16		24		30		30		22		21		N/A	
<i>DSM-5 Oriented Scales</i>																				
Depressive Problems	1	N	N/A		9	C	5	N	5	N	5	N	11	C	12	C	8	C	N/A	
Anxiety Problems	6	N	N/A		6	N	5	N	7	N	14	C	13	C	15	C	9	C	N/A	
ASD Problems	10	C	N/A		12	C	10	C	11	C	10	C	15	C	15	C	12	C	N/A	
ADHD Problems	9	N	N/A		8	N	5	N	12	C	9	N	10	B	7	N	11	C	N/A	
Oppositional Defiance	2	N	N/A		5	N	2	N	6	N	3	N	8	B	8	B	9	C	N/A	
Internalising Behaviours	61	B	N/A		66	C	51	N	60	B	70	C	78	C	82	C	72	C	N/A	
Externalising Behaviours	61	B	N/A		58	N	48	N	69	C	61	B	74	C	64	C	67	C	N/A	
Total Score	61	B	N/A		64	C	56	N	69	C	72	C	82	C	77	C	72	C	N/A	

(Note: R= Range; N=Normal; B=Borderline; C=Clinical)

Matt. Before intervention, Matt's parents gave him a borderline range score of 61 for each of the internalising behaviour, externalizing behaviour, and total behaviour problem domains. They rated him to be in the clinical range for withdrawal and ASD problems. His scores for all other scales were in the normal range, including sleep problems. No post-treatment data was collected for Matt.

Catherine. Catherine had pre-intervention scores within the clinical range for internalising behaviours, externalizing behaviours, and total behaviour problems, as well as all DSM-5 oriented scales. She was also rated in the clinical range for withdrawal and attention problems. His parents gave him scores in the borderline range for the emotionally reactive, somatic complaints, sleep problems and aggressive behaviour domains, and a score in the normal range for the anxious/depressed domain. No post-treatment data has yet been collected for Catherine.

Andrew. For Andrew, his parents scored him in the normal or clinical range for all domains pre- and post-intervention. Following intervention, reductions were seen in all domains, except sleep problems which remained constant and within the normal range. Most markedly, his total problem behaviour score, as well as his internalising behaviour and depressive symptoms scale scores all decreased from the clinical to normal range. These results suggest an improvement in Andrew's behaviour and emotional problems from pre- to post-intervention.

George. For George, his mother scored him in the clinical range for total problems at both pre- and post-intervention. Following intervention, reductions were seen in his externalizing behaviour score (69 down to 61), moving him from the clinical to borderline range. Reductions were also seen in three of the empirically based scales. His withdrawal scale score moved from the clinical to

normal range (6 to 4), and his aggressive behaviour score moved from the borderline to normal range (21 to 15), while reduction in his attention problems remained in the clinical range (9 to 7). Increases were seen in the scores of the remaining four empirically based scales. His emotionally reactive, and anxious/depressive scale scores moved from the normal to borderline range (both 3 to 7), and his somatic complaints score moved from the normal to clinical range (2 to 7). Interestingly, his sleep problem score also moved from the normal to borderline range (7 to 8). Increased on the DSM-oriented scales were reported on the anxiety problem subscale, moving from the normal to clinical range (7 to 14). George's depressive problem score remained constant and within the normal range (5). Reductions were seen in his ASD problem scores, but this remained within the clinical range (11 down to 10). His ADHD score reduced from the clinical to normal range (12 down to 9), and his oppositional defiance score decreased and remained within the normal range (6 down to 3). Overall, mixed results were reported for George in comparison to baseline.

Harry. For Harry, his parents scored him in the borderline or clinical range for all domains pre-intervention. Following intervention, reductions were seen for his total behaviour problem score (82 to 77) and externalizing behaviour score (74 to 64), but these remained within the clinical range. An increase was seen in his internalising behaviour score (78 to 82), which also remained within the clinical range. Reductions were seen in four of the empirically based scales, with sleep problems, attention problems and aggressive behaviours moving from the clinical to normal range (scores of 10 to 7, 8 to 5, and 26 to 20 respectively). Increases were seen in four of the empirically based scale scores, with somatic complaints moving from the borderline to clinical range (6 to 8), and emotionally reactive

problems, anxious/depressed problems, and withdrawal problems remaining in the clinical range (15 to 16, 10 to 11, and 7 to 9 respectively). Harry's ASD problem scores remained constant and within the clinical range, suggesting the CBCL was unable to detect any changes in his ASD symptomatology. No changes were seen in his oppositional defiance score (8, borderline range). However, his ADHD problem score moved from the borderline to normal range (10 to 7). Small increases were seen in his depressive problems and anxiety problems scores, remaining within the clinical range (11 to 12, and 13 to 15 respectively). Overall, mixed results were reported in comparison to baseline.

Treatment Acceptability

TARF-R. Post-intervention results of the TARF-R for Andrew, George and Harry's parents are reported in Table 11. Overall, for the families that completed intervention, parents generally reported the interventions to be acceptable, effective, and clear to understand, but perceived the interventions to require a great deal of effort and time to implement.

Table 11. Post-Intervention Treatment Acceptability Scores from TARF-R for Study 2 Participants

Variable scores	Andrew		George		Harry		Max score
	Mother	Father	Mother	Father	Mother	Father	
Total Acceptability	103	87	110	N/A	100	109	121
Reasonableness	20	18	21	N/A	18	21	21
Willingness	19	17	20	N/A	18	20	21
Effectiveness	18	19	19	N/A	18	20	21
Cost	14	11	14	N/A	14	14	14
Negative Side-Effects	16	15	20	N/A	17	20	21
Disruption/Time	7	11	8	N/A	15	14	21
Problem Severity [°]	12	12	8	N/A	10	9	14
Understanding of Treatment [°]	6	5	7	N/A	7	7	7

[°] = not included in the Total Acceptability score

Andrew. For Andrew, his mother showed a high-level of treatment acceptability overall (103/121), whereas his father reported a more moderate level of overall treatment acceptability (87/121). Results suggest that both of Andrew's parents were willing to carry out the intervention (mother: 19/21, father: 17/21), and they perceived the type of treatment to be reasonable (mother: 20/21, father: 18/21), effective (mother: 18/21, father:19/21), and very affordable (both: 14/14). Results suggest that both of his parents believed the intervention to have some negative side effects (mother: 16/21, father: 15/21), and that it caused a high-level of disruption, and required extensive time to implement (mother: 7/21, father: 11/21), but that both parents were relatively clear in their understanding of the treatment (mother: 6/7, father: 5/7). Both

parents perceived Andrew to have high sleep behavior problems in comparison to same aged peers following intervention (both: 12/14).

George. For George, his mother showed a high-level of overall treatment acceptability (110/121). Results suggest she was very willing to carry out the intervention (20/21), and that she perceived treatment to be very reasonable (21/21), effective (19/21), very affordable (14/14), and with few negative side effects (20/21). However, results suggest that she perceived the intervention to cause a high level of a disruption and required a great deal of time to implement (8/21), but that she was very clear in her understanding of the treatment (7/7). Following intervention, George's mother perceived him to have moderate sleep behavior problems in comparison to same aged peers (8/14).

Harry. For Harry, results showed a high level of treatment acceptability overall with a score of 100/121 and 109/121 for his mother and father respectively. Results suggest that both of Harry's parents were willing to carry out the intervention (mother: 18/21, father: 20/21), and they perceived the type of treatment to be reasonable (mother: 18/21, father: 21/21), highly effective (mother: 18/21, father: 20/21), affordable (both parents: 14/14), and have few undesirable side-effects (mother: 17/21, father: 20/21). Results did suggest that his parents perceived the intervention to cause a reasonable level of disruption and time to implement (mother: 15/21, father: 14/21), but that both parents were very clear in their understanding of the treatment (both parents: 7/7). Both parents perceived Harry to have moderate sleep behaviour problems in comparison to same aged peers following intervention, with a score of 10/14 and 9/14 for Harry's mother and father respectively.

Post-treatment discussions.

Andrew. Andrew's mother stated that they were satisfied with the intervention process and that all of the treatment suggestions made sense. She felt supported by the researcher, and appreciated that daily contact meant immediate modifications to Andrew's treatment plan. She said that she felt she had not got as much success from the treatment as she had hoped for, in that Andrew was still having frequent night wakings, and although his wake time had extended, it had not reached the level she had hoped for. However, she felt that Andrew was "pushed to the limit of his capabilities". She felt that the improvements she did see in his independence, night wakings and later morning wakings was attributed to her consistency and repetition, and pushing his bedtime back to apply sleep pressure. She said that she felt the intervention had impacted on her personally, as she was less tired during the day due to getting more sleep. Andrew's mother did not notice any changes in his daytime behaviours, and said that he still appeared to be tired during the day.

George. George's mother stated that she was very satisfied with the intervention process and the significant improvements George made with regards to his independence at sleep onset and following night wakings, shorter sleep onset latency, and later wake times. George's bedtime had shifted later during the intervention, but his mother felt that her self-efficacy around implementing sleep behaviour strategies had improved to a level that she was confident resolving this herself. She believed that the social stories and gro clock had the greatest impact on his sleep behaviours as they aided his comprehension. She also believed that being made more conscious about her own actions, and changing her behaviours so she was giving him less attention and being more

consistent was pivotal in changing his sleep behaviours. She said that treatment resulted in significant improvement in her sleep quality and quantity, making her feel better during the day. George's mother noticed some changes in his daytime behaviours, stating that his comprehension and speech had "rocketed" since he started intervention, and that he stopped the constant need to squeeze hands during the day. However, she was unsure as to whether these changes were a result of the intervention, or coincidental. In addition, the intervention made her realise that he was capable of understanding more than she had thought, and that social stories could be a valuable tool for other aspects of George's development.

Harry. Harry's parents stated that they were very satisfied with the overall treatment process, and appreciated that it was "direct and straightforward... simple, short and clear". They said that initially the intervention was challenging as it was intensive, requiring a lot of "work and effort", but that as their own sleep improved, implementing the treatment became more manageable. Harry's father believed that the researchers availability to discuss his concerns and queries was invaluable. They felt that they had learnt that they did not need to be "forceful and reprimanding" to have an impact on his sleep behaviours. They felt that the reinforcement procedure made the biggest difference to Harry's behaviours, as he had an incentive to go to sleep. Both of Harry's parents felt that their own sleep had improved and that they are now much calmer and able to react more positively in response to Harry's challenging behaviours. In addition, they felt that Harry's challenging behaviours prior to bedtime had decreased in intensity since intervention, and that he is more accepting of his parents instructions about getting ready for bed, on some nights showing no resistance at all.

Chapter 7

Discussion

Research Questions

The current study had five research aims: 1) to investigate the use of FBA to inform interventions for sleep problems in children with ASD; 2) to examine the effectiveness of individualised treatments for sleep problems, including co-sleeping; 3) to examine the effect of successful treatments on daytime behaviours and ASD symptomatology; 4) to examine the effect of successful treatments on parent well-being; and 5) to evaluate parent acceptability and understanding of the assessment and treatment process.

Study findings

In response to treatment, sleep onset latency reduced for all children, except Matt who withdrew from the study. The frequency of curtain calls was reduced for four of the six children, and for one child they returned to baseline levels. For Matt, the frequency of curtain calls increased following baseline. There was a reduction in the frequency of night wakings for all children except Matt, and Catherine who had not yet completed intervention. A reduction in the duration of night wakings was also seen for all children, except Catherine. Parental presence during sleep onset was eliminated for all six children, and co-sleeping following night waking was eliminated for all children except Matt.

Following intervention, two out of three parents reported decreases in total sleep difficulties from above average to average ranges on the CSHQ, with one child having a slight increase in total difficulties.

DASS-21, results indicated that scores on the depression index either improved or remained constant for the parents of all four children who completed intervention, whereas anxiety and stress scores were more variable. All parents of the three families in Study 2 that completed intervention reported improvements in their sleep quality following intervention, as reported on the PSQI. Mixed results were found regarding changes to relationship quality for the two families who completed the RQI pre- and post-intervention.

All three families who completed the CBCL pre- and post-intervention reported decreases in their child's externalising behaviour scores, as well as improvements in the attention, aggression, and ADHD characteristics domains. Mixed results were seen in changes of total behaviour difficulties, and internalising behaviours. Following intervention, all three children who completed the GARS-3 were perceived to have improvements in their overall ASD symptomatology, in particular their restricted/repetitive behaviours.

The post-treatment interview and results of the TARF suggested that for the four families that completed intervention, parents were satisfied with, and had a good understanding of the intervention but also felt that implementing an intervention required significant amounts of time and levels of disruption to the family resulting in some negative side-effects.

The utility of FBA to inform intervention. In the current study, the outcomes of the FBA process were used to develop individualised and

comprehensive treatments for each child. FBA has a strong evidence-base for informing interventions for challenging behaviours in children with ASD (Campbell, 2003; Hanley et al., 2004; Hansen & Wadsworth, 2015). However, few studies have documented the use of FBA in order to develop interventions for sleep problems (Didden et al., 2002; Hanley et al., 2014; Jin et al., 2013), and only four studies were found that used FBA when treating co-sleeping as one target of their intervention (Didden et al., 2002. Moore 2004; Weiskopp et al., 2001; Weiskopp et al., 2005). Using FBA to inform interventions for co-sleeping was considered important in the current study, as interventions for challenging behaviour that are based on FBA have been found to be more effective than interventions that are not (Beavers et al., 2013; Hanley, 2016; Spruyt & Curfs, 2015). The current study built on existing research by investigating the effectiveness of FBA to inform interventions for children who have sleep problems that include co-sleeping.

A key finding of this study was that the FBA process helped to isolate and identify the unique antecedent and consequence variables maintaining the sleep problems for each child. Although the sleep problems and the topography of the sleep interfering behaviours were very similar between children, FBA revealed that the variables impacting on the behaviour were not. For example, variables such as access to tangible items during bedtime or in the morning, and lack of physiological sleep pressure uniquely contributed toward the sleep problem for each child. This finding emphasises the need for treatments to be individualised so that the specific variables maintaining their sleep problems are targeted. This would enhance the likely effectiveness of interventions as, unlike generically applied strategies, they will directly target the unique factors contributing toward

the sleep problem (Beavers et al., 2013; Hanley, 2016; Spruyt & Curfs, 2015). This is in line with recent research suggesting there are limitations to a 'one size fits all' solution to sleep problems, and that interventions need to be tailored to the individual (Singh & Zimmerman, 2015. Spruyt & Curfs, 2015). Furthermore, this study provides support for existing research that suggests that sleep problems are multifactorial requiring multimodal treatments (Singh & Zimmerman, 2015; Spruyt & Curfs, 2015).

The effect of selected treatments on sleep-related outcomes. In this study, individual treatment approaches were selected based upon the outcomes of FBA, current and emerging evidence-based treatments for sleep problems in typically developing children and children with ASD, evidence-based treatments for challenging behaviours other than sleep problems, and collaboration with parents. Treatments included sleep hygiene practices, modification of the bedroom environment, visual supports, stimulus substitution, faded bedtime, camping out procedures, extinction, reinforcement procedures, and medication. The effect of each of these treatments is given separate consideration below.

Modification of the bedroom environment. Harry was the only child for whom the bedroom environment required modification. This included removing access to his toys (finished box), and moving bedrooms to one further away from the lounge. Removing access to toys, and parent interaction in the nearby room appeared to eliminate the social and tangible reinforcers that were contributing toward the sleep problem for Harry. These procedures, in conjunction with planned ignoring, resulted in reduced sleep onset latency. This finding is consistent with previous research that demonstrates that small adjustments to

the sleep environment can have positive effects on sleep behaviours (Christodulu & Durand, 2004; Singh & Zimmerman, 2015; Tan et al., 2012).

Visual supports.

Social stories. Social stories were used to help aid the child's understanding of the changes made under intervention. Social stories were used by all children in study 2, and appeared to have a positive impact on their sleep outcomes. In line with the current research, social stories were found to be easily individualised for each child, and were easily modified by parents to reflect changes to the treatment plan (Bozkurt & Vuran, 2014; Fray, 2010; Singh & Zimmerman, 2015). Social stories have been used effectively as part of multicomponent treatment packages to teach children with ASD a variety of skills, including coping with grief and eating difficulties (Moore, 2004). Only one other study was identified which used social stories as a component of treatment for sleeping problems in children with ASD (Moore, 2004). The findings of this study are consistent with those of Moore (2004) that also demonstrated improvements in co-sleeping, sleep onset latencies and night wakings. It is unlikely that social stories alone resulted in improved sleep outcomes (Bozkurt & Vuran, 2014; Styles, 2011; Test et al., 2011), but they did appear to be a socially valid complementary tool for informing the children in this study about changes in expectations. Social stories were also well liked by both parents and children. George's mother specifically noted during her post treatment interview that she believed social stories were key to George's progress, due to it aiding his comprehension about what was expected of him.

Groclocks. Groclocks were used by five children in the current study to help them to learn the distinction between sleep and wake time. No improvements have been seen in the frequency of night wakings or early waking behaviours in Catherine's intervention so far, however the frequency of night wakings decreased for Ben, Harry, Andrew and George, with George also showing marked improvements in his ability to stay in bed past 6am. This suggests that, for these three children, Groclocks were an effective tool to teach the children when they were able to get out of bed. This study appears to be the first to investigate the use of Groclocks as a component of sleep interventions.

Video model. Ben used a video model. As with social stories, it is difficult to determine whether video modeling alone was sufficient to result in behaviour change. However, all of Ben's targeted behaviours improved as a result of intervention, suggesting that video modeling at the very least may have supported Ben to understand the changes to his sleep routine. Video modeling has been used to teach a range of behaviours to children with ASD, for example perspective taking (Bellini & Akullian, 2007) and asking for preferred objects (Banda, Copple, Koul, Sancibrian, & Bagschutz, 2010), but the current study is the first known study to use video modeling to treat sleep problems in a child with ASD.

Visual symbols. Visual symbols were used on Ben's parents' door as another tool to aid learning the distinction between sleep and wake time. As all of his sleep behaviours improved, it is difficult to isolate the importance of this visual symbol in scaffolding his understanding. However, this finding is in line with previous research that has found similar reductions in sleep difficulties

when using visual symbols as a component of intervention (Moore, 2004; Reed et al., 2009; Weiskopp et al., 2001; Weiskopp et al., 2005).

Sleep hygiene. Sleep hygiene was addressed as a component of intervention for four of the participants in this study. This included the elimination of day sleeps for Matt, Catherine and Ben, and incorporating toileting into the bedtime routine and a consistent wake time for Harry. Previous research suggests that establishing positive sleep hygiene is an essential first step in sleep interventions, and is something that should be addressed in order to increase the likely effectiveness of other components of intervention (Johnson et al., 2009; Vriend et al., 2011). All of the children in the study who addressed sleep hygiene, with the exception of Matt, showed improvements in bedtime resistance during their intervention, and all had a reduced sleep onset latency, which may have been helped by implementing good sleep hygiene practices (Spruyt & Curfs, 2015).

Faded bedtime. Faded bedtime was a procedure recommended for use with Harry, Andrew and Matt. Fading bedtime did not appear to have an effect on sleep onset latency or wake time for Matt or Andrew. However, for Matt, a sudden reduction in sleep onset latency was observed when his bedtime was faded back for the second time. A faded bedtime procedure appeared to have a positive effect on Harry's sleep onset latency, with immediate reductions upon the introduction of intervention. A PERB (in which sleep onset latency temporarily increased) was observed when Harry's bedtime was faded forward, suggesting that sleep onset latency was effected by this change. For Matt and Andrew, it is possible that fading bedtime did not result in changes in sleep onset latency or wake times, due to the elimination of co-sleeping during sleep onset for these two children having

a confounding effect on these outcomes. By contrast, Harry's parents were never present during sleep onset, and therefore fading bedtime may have had a more significant and pertinent effect. Overall, the use of a faded bedtime without response cost procedure in this study appeared effective when parent presence during sleep onset did not need to be addressed, and adds to the literature that uses it as a component to improve sleep initiation problems in children with ASD (for example, Johnson et al., 2013; Papadopulus et al., 2015).

Stimulus substitution. A stimulus substitute was used to provide a nonsocial and consistent discriminative stimulus for sleep that replaced and mimicked the reinforcement provided by parents. Stimulus substitution was used as a component of intervention for Ben, George, Harry, and Catherine to target the need for parent presence during sleep onset, co-sleeping, sleep onset latency, frequency of curtain calls, and night wakings.

Introduction of a sleep item. For Ben and George, stimulus substitution included the use of the 'body' and a sensory ball respectively. These treatments appeared effective, as for Ben and George, parent presence and co-sleeping was eliminated, and sleep onset, frequency of curtain calls, and night wakings improved.

Music. For Harry and Catherine, stimulus substitution included soft music being played in their rooms during sleep onset and throughout the night. For Harry, co-sleeping was eliminated, and improvements were seen in his sleep onset latency, and frequency of curtain calls and night wakings. Catherine had eliminated all parental presence and her sleep onset latency had improved, but positive effects on her frequency of curtain calls and night wakings had not yet occurred.

Use of a stimulus substitute was deemed socially valid and easy to use by these families, with Ben's mother stating that she believed the 'body' had the biggest impact on Ben's improvements, and that he adored his 'body'. Using a stimulus substitute during intervention for these children built on previous research that suggests correlations between sensory modulation difficulties and sleep problems in children with ASD (Reynolds et al., 2011), and that a comprehensive understanding of the motivations for behaviours and the specific sensory qualities that are being reinforced is required to decrease or eliminate these behaviors (Joosten et al., 2009; Patel et al., 2000). There is a lack of literature that uses stimulus substitution to aid sleep in children with ASD, and therefore this study, that showed improvements in sleep behaviours with the use of sleep items, is extending what is known in this field.

Camping out. Camping out procedures are used as a more gentle alternative to standard extinction to remove parents presence during sleep onset and following a night waking. A camping out program was used for Matt, Andrew, George and Catherine.

Using this procedure instead of a standard extinction procedure was decided on in collaboration with the parents, as it was hypothesised to lead to less distress for both the children and parents. For Andrew and George, intervention appeared to successfully reduce the frequency and duration of night waking. Parental presence was removed during sleep onset, and co-sleeping during the night, as each of these children learnt to reliably sleep independently through the night in their own beds. Although Matt and Catherine did not complete intervention, by the end of their involvement in the study, they no longer required a parent's presence to settle to sleep at sleep onset. Co-sleeping

was eliminated for Catherine, and the frequency with which Matt co-slept during the night had also decreased since baseline, but had not been extinguished.

Howlin (1984) was the only study found that used a camping out procedure to address sleep problems in a child with ASD. For the two children who completed intervention in the current study, co-sleeping was eliminated, and both night wakings and sleep onset latencies decreased, which is consistent with the findings of Howlin's (1984) study. The current study appears to be the first to extend knowledge on the use of a camping out procedure with children with ASD in 33 years.

PERB's were observed for Matt, Catherine, Andrew and George when their parents' presence was faded. Due to the study design and the fact that parent fading was done in conjunction with Vallergan for George, it is hard to tell whether these reactions were of lesser intensity than if standard extinction had been used. Past research has suggested that graduated extinction is favourable to standard extinction, as it may reduce the intensity of the PERB (France & Blampied, 2005) and it has higher social acceptability and adherence levels (Lerman et al., 1999; Vriend et al., 2011).

Standard extinction procedures. An extinction procedure was used with two children included in this study (Harry and Ben). Results for Harry showed that standard extinction lead to the successful and immediate extinction of co-sleeping, immediate decrease in sleep onset latency, and a reduction and then elimination of curtain calls and night wakings. For Ben, his sleep onset latency and frequency of curtain calls were immediately reduced, and co-sleeping was reduced and then eventually eliminated, and his frequency and duration of night wakings was reduced. This finding is consistent with existing research that

suggest standard extinction has a rapid and positive effect on bedtime resistance and night wakings (Didden et al., 2002; Wolf et al., 1964) as well as co-sleeping (Weiskopp et al., 2005).

Overall, these families were able to adhere to an extinction procedure, successfully removing the reinforcement that was maintaining the unwanted behaviors. The use of extinction procedures in the current study provides support for the body of literature that shows extinction procedures can effectively reduce problematic sleep behaviours, including co-sleeping, in children with ASD (Vriend et al., 2011).

France and Blampied (2005) noticed that standard extinction procedures resulted in the greatest PERB, but one of the quickest resolution of problematic behaviours. PERBs did not occur across all sleep outcomes following the use of extinction procedures, for example no PERB was evident for Harry in relation to his sleep onset latency. For Oliver, his sleep problems were resolved more quickly than the other children. However, Ben's problems took a long time to resolve. PERBs were not reported to be problematic. It is likely however that PERBs did occur. In particular, Harry displayed spikes in frequency of curtain calls and Ben also showed a clear increase in his frequency of night wakings following the introduction of extinction. It is possible that the PERBs that occurred were not problematic for several reasons. First, they occurred in conjunction with other intervention components that may have lessened their impact, for example Harry's faded bedtime and Ben's 'body'. Second, parents were given psycho-education about the occurrence of PERBs, and therefore it is possible that they perceived them to be normal and not problematic. A third possible explanation is that social stories scaffolded their understanding of what

was occurring. It is also possible that the eliminated target was not a primary factor maintaining the behaviours. Furthermore, it is possible that the research did not capture the impacted behaviors, for example extinction may have resulted in an increase in temper tantrums during the bedtime routine that was not measured.

Reinforcement procedures. Reinforcement was used for all children in the study to reinforce the occurrence of target behaviours. Reinforcement was in the form of social praise and tangible rewards given immediately after the child woke in the morning. In all six cases, reinforcement appeared to be well liked by both parents and children, was easy to implement, and appeared to successfully reinforce the children's learning of desired behaviours. In fact, Harry's parents believed that the reinforcement procedure had the most impact on his behaviors, as it provided him with an incentive to go to sleep at night. The use of reinforcement in this study adds to the body of literature that uses this method to shape sleep related behaviours in both typically developing children and children with ASD (Johnson et al., 2013; Knight & Johnson, 2014; Moon et al., 2011).

Pharmacological interventions. Medication is commonly used in sleep interventions, as it is easy to use and has immediate effects (Richdale, 2013). Medication was used for five of the six children in the study. Matt and Ben were regularly taking melatonin before the study commenced, and used it throughout intervention. Before involvement in the research, Andrew had a prescription for melatonin, and George had a prescription for Vallergran, which they used sporadically.

Working in collaboration with their doctors, both George and Catherine used Vallergran as a component of their sleep interventions. For George, his sleep

onset latency and curtain calls improved with intervention, and were maintained after Vallergan was faded out, suggesting that learning of behaviour strategies occurred. Due to the study design, it is hard to tell if Vallergan had any impact on his level of distress, but it is likely that PERBS were reduced as a result. Initial results for Catherine suggest that Vallergan had a possible effect on her sleep onset latency and frequency of curtain calls, as these outcomes progressively improved after the introduction of Vallergan, despite her having no parent presence. No noticeable impact was observed on her night wakings. The positive effects of Vallergan medications in conjunction with behavioral interventions in this study adds support to the literature which shows combining pharmacological interventions with behavioral interventions can be an effective part of sleep intervention for children (France et al., 1991; Selim et al., 2006).

Outliers, anomalies and interesting observations.

Ben. After a peak in frequency of curtain calls on the first day of intervention, these rapidly declined and were eliminated for the remainder of the intervention. The exceptions to this progress in frequency of curtain calls were on days where he had day naps, suggesting that sleep pressure was an important factor for Ben that impacted on his sleep initiation problems. As soon as co-sleeping during the night was targeted through extinction in phase two, Ben did not co-sleep throughout the intervention, with the exception of five nights. On the majority of these nights, circumstances were not typical for Ben (i.e., his grandfather rather than his parents was involved in his nighttime routine, or he had a nightmare), which highlights Ben's reliance on consistency in routines for positive sleep behaviours.

Results of the CSHQ indicated that his parents perceived improvements in his bedtime resistance, but no changes in his sleep onset latency or night wakings, despite the sleep diaries demonstrating improvements in these areas. There are several possible reasons for the inconsistency between these two measures. The CSHQ and sleep diaries measure two different constructs, and therefore differences are likely. It is possible that the CSHQ was not able to accurately capture changes in sleep outcomes. It is also possible that Ben's parents were more aware of his sleep behaviours following their involvement in the research in comparison to pre-intervention, and their scores reflected this.

Matt. Matt had consistently low sleep onset latencies and no curtain calls during baseline. This is most likely due to his melatonin medication influencing sleep pressure, and his mothers' presence decreasing his need to actively attain attention. His SOL and frequency of curtain calls increased significantly when his mother moved out of the door but was still visible and then decreased to baseline levels four days later, indicating the presence of a PERB response. No PERBs were observed as his mother moved to increasingly less visibility from Matt. This suggests that it was his mother's proximity, rather than her contact or visibility, that Matt associated with sleep, as disrupting this association lead to a temporary increase in sleep resistant behaviours. His frequency of night wakings returned to baseline levels during the last 1½ weeks of intervention, and his duration of night wakings were minimal during this time. This is most likely a consequence of his parents allowing him to co-sleep frequently during this period. Matt was more likely to seek out his parents following a night waking if he was intermittently reinforced with co-sleeping, and his duration of night wakings was likely to be low as co-sleeping meant he was immediately reinforced with his parents

attention and did not need to engage in sleep interfering behaviors to acquire it. His goal of waking after 6 am also became more difficult for Matt to attain as intervention progressed. It is possible that his sleep was consolidating, resulting in a period of earlier wake times. It is also possible that his parents had unrealistic expectations around his sleep duration, as his early bedtime was not conducive to him sleeping later.

There are several reasons why Matt may not have completed intervention. His parents cited that other children had begun to disrupt their sleep and they were too tired to continue. It is also possible that other extraneous factors contributed to their decision to withdraw, or that this particular intervention was simply too difficult to carry out for this family at this point of time. Matt was the youngest participant in the study, so it is possible that his age was also a barrier to his interventions success.

Catherine. A peak in Catherine's sleep onset latency and frequency of curtain calls was apparent on the third day of intervention, at which time resistance about having no bottle in bed was reported. This rapidly declined when milk was completely removed from the bedtime routine. This resistance followed by a sudden decrease in problem behaviors at sleep onset suggests that her milk bottle was a factor in maintaining her sleep interfering behaviors. Another peak in sleep onset latency and curtain calls was apparent when Catherine's mother moved to be beside the door, and more resistance occurred when all parent presence was removed, suggesting that her mother's physical rather than visual presence was helping to maintain her sleep interfering behaviours at sleep onset.

Her parents goals of her having no parent presence or bottle to settle, and no co-sleeping were met as soon as these factors were removed according to the treatment regime, suggesting that Catherine's family were able to adhere to the corresponding intervention components, and that they were effective for achieving these goals. Her final goal of waking after 6am became more variable during intervention in comparison to baseline, suggesting that intervention can disrupt wake times.

Andrew. In general, not as much improvement was seen in Andrew's sleep behaviors as was desired. Results suggest that his mother's proximity rather than visual presence was helping to maintain his bedtime resistant behaviors, as peaks in his sleep onset latency and curtain calls were observed when she moved to the door and then again when she moved out to the hallway, and no PERBS were observed when she moved out of sight. In addition, it appears that sleep pressure was also a contributing factor to his sleep difficulties, as moving his bedtime later impacted positively on his sleep onset latency and frequency of curtain calls.

His goal of no co-sleeping was consistently met following the introduction of intervention. As he was still having night wakings this suggests that the camp out procedure was effective and able to be implemented consistently. The goal of waking up to 6am remained variable, and this may be a result of his sleep consolidating, or because Andrew may have melatonin/circadian rhythms that are not behaviourally operational.

Andrew's results on the CSHQ mostly reflect his performance seen in sleep diaries. Most domains remained unchanged. Small increases were seen in reported bedtime resistance, sleep anxiety and daytime sleepiness. It is possible that bedtime resistance and sleep anxiety reflect his variability in sleep onset

latency and curtain calls, and possible concerns about settling independently. Although improvements were seen in night wakings, this was not reflected on the CSHQ, possibly as his goals were not completely achieved.

George. During the later stages of phase three, there was a period of increased SOL and curtain calls. It is possible that these were a side effect of feeling better following a period of illness, in which he did co-sleep on one occasion. Consistent improvements in all sleep outcomes measured were observed after the introduction of phase four, in which his mother was no longer visible to him. A possible PERB was displayed in his number of curtain calls, which rapidly diminished. This suggests that it was his mothers' visual presence, rather than her physical proximity, that was maintaining his sleep problems.

George's mother reported an increase in parasomnias, in particular nightmares, following intervention. This may have been a side effect of the intervention, which can possibly be explained by his sleep consolidating and therefore resulting in more periods of REM sleep, and consequently more nightmares. It is also possible that as he aged throughout the intervention, his imagination developed, resulting in more nightmares. George's mother herself suggested that his imaginative play during the day had improved throughout the time he was involved in the study.

Harry. Harry's parents goals of him sleeping independently during the night and having a sleep onset latency under 30 minutes was attained throughout intervention with the exception of two and four nights respectively, and his goal of no night wakings was achieved during the last half of intervention with the exception of four nights. Most of these times in which he did not obtain his goals, he had either had a day nap, or his grandparents were babysitting, suggesting

that sleep pressure and changes in his social environment impact on his sleep behaviours, and that routine and consistency are important for him to achieve the desired outcomes. On the other two nights when he did not achieve his SOL goal, he was within five minutes of his goal.

Harry's night wakings score on the CSHQ decreased, but remained at above average levels, despite diaries showing infrequent night wakings in the last half of intervention. This is likely due to the diaries and CSHQ measuring different constructs. Sleep diaries report a night waking only if the child purposefully disturbs their parents, whereas the CSHQ captures night wakings that the parents are aware of, but do not result in their child disrupting them. Harry's parents reported that they were aware that he frequently left his room during the night and spent time in the bathroom, without entering their room, and their score likely reflects this. In addition, no changes were reported in his sleep onset latency by his parents on the CSHQ, despite the dramatic decreases evident in his sleep diaries. It is possible that the CSHQ was not able to accurately capture these changes.

The impact of successful treatment on daytime behaviours and ASD symptomatology. Four of the children in this study successfully completed sleep interventions. Evidence supplied by these parents in the post-treatment interviews suggested that three out of four of the families perceived changes in their children's daytime behaviours after intervention, which included increased attention and engagement at school, an improvement in comprehension and speech, a decrease in self-stimulatory behaviours, and a decrease in intensity of evening temper tantrums. These perceived improvements were also reflected in

CBCL and GARS scores for the three children in the study who completed intervention.

CBCL. Before intervention, all five children in study 2 had total CBCL scores and internalising behaviour scores in the borderline or clinical ranges, and four children had externalising behaviour scores in the borderline or clinical ranges. Parent's scores for Andrew, George and Harry demonstrated mixed results for changes in total behaviour difficulties and internalising behaviours. However, all three children had decreases in their externalising behaviour scores following intervention. In addition, scores suggested that all three children had improvements in their attention and aggressive behaviours, and decreases in ADHD characteristics. This is in line with Personen et al. (2010) who found correlations between sleep problems and attention deficits and externalising behaviours in typically developing children.

Of note is that scores on the sleep problem scale of the CBCL were mixed, with Andrew's score being unchanged, George's increasing, and Harry's decreasing. This suggests that the CBCL was either unable to accurately detect changes in sleep behaviours, or it measured different constructs to the current studies sleep outcomes.

While most recent studies exploring sleep and challenging behaviours in children with ASD are correlational, demonstrating a strong link between sleep difficulties and daytime behaviour problems (for example Sikora et al., 2012), this relationship is not necessarily causal, and may be bidirectional (i.e., sleep problems may result in more challenging behaviours, and challenging behaviours may result in sleep problems). It is important that future research is conducted in order to understand this relationship. Only three case-studies were found in the

literature that investigated the effect of sleep interventions on the daytime behaviours of children with ASD, and all demonstrated positive changes (Malow et al., 2006; Moon et al., 2011; Reed et al., 2009). The current study adds to this literature, demonstrating the promising effect of sleep interventions on daytime behaviours.

There are several possible reasons why improvements were observed in children's daytime behaviours following sleep interventions. For example, it is possible that challenging behaviours and sleep problems have similar origins and maintaining factors within the individual, and therefore changing the factors that influence one problem can have an impact on other problem behaviours. It is also possible that sleep deprivation may present in the form of challenging behaviours. In addition, parents gain parenting skills through sleep interventions that may generalise to and be used in other situations. Furthermore, sleep interventions that result in parents getting more sleep may lead to them feeling more relaxed during the day and therefore having more rational and objective reactions to their child's behaviours, which is reflected in the CBCL scores.

GARS-3. In general, results of the GARS were positive, with all three children demonstrating improvements in their overall ASD symptomatology. All subscales had mixed results across the children, with the exception of the restricted/repetitive behaviour subscale, where all three children showed improvements. This is in line with Reed et al. (2009) and Shochat et al. (2000), who found that improvements in sleep lead to a decrease in repetitive/restrictive behaviours. With improvements in restricted/repetitive behaviours seen following improvements in sleep behaviours, the current study suggests that there may be a bidirectional relationship between these two factors, with sleep

problems possibly contributing to sensory seeking behaviours. Limited research has been conducted on the relationship between sleep and ASD symptomatology, with most studies being correlational. Only one study was found that collected quantitative data on changes in ASD symptoms following sleep improvements (Reed et al., 2009), therefore this study adds to the new research field, and suggest promising effects of sleep interventions on ASD symptomatology.

The effectiveness of successful treatments on parent wellbeing. A recurring theme in all post treatment interviews was that the parents of the children who successfully completed the intervention perceived themselves to be less tired following treatment than before intervention. In addition, Harry's parents reported that the intervention resulted in them feeling calmer. Another positive impact of the intervention was the increased self-efficacy and confidence to tackle future sleep problems that was reported by Ben's parents and George's mother. This is in line with the findings of Weiskop et al. (2005) that also had reports of increases in parent's confidence and self-efficacy as a result of sleep treatment. These anecdotal reports were partially supported by the results of the DASS-21, PSQI and RQI.

DASS-21. Before intervention, all parents had depression, anxiety, and stress scores in the normal range, with the exception of George's mother. This is in contrast to the findings of previous research that suggests parents of children with ASD and sleep problems are at a higher risk of being clinically depressed (Foody et al., 2014; Meltzer, 2011; Tilford et al., 2015), anxious, and stressed (Doo & Wing, 2006; Hoffman et al., 2008; McStay et al., 2014; Meltzer & Mindell, 2007). Changes in depression, anxiety, and stress pre- and post-intervention

were mixed. All parent's depression scores either decreased or remained constant following intervention, but anxiety and stress scores were more variable. Of importance, is that George's mothers scores on all variables decreased significantly between the two time points, and were all in the normal range as a result. Although the DASS-21 is not able to recognise reasons for change (Moore, 2004), it is possible that depression scores decreased as a result of the parents getting better quantity and quality of sleep, and/or the child's behaviours improving. However, it is likely that extraneous factors had an important role on DASS-21 outcomes, making it difficult to directly attribute changes to the results of the study. Harry's mothers anxiety scores increased from the normal to mild range following intervention. These changes may be attributed to the sleep intervention, or may be due to other factors, for example concerns about Harry starting school after the completion of the intervention. Both of Ben's parent's anxiety scores also increased slightly following intervention. One reason for this could be related to a concern they reported about being anxious about what Ben was doing if he was not sleeping with them. All studies found that investigated the relationship between sleep problems in children with ASD and parent well being were cross-sectional studies, therefore this is the first known study to demonstrate changes in parents wellbeing as a result of sleep interventions.

PSQI. Overall, results of the PSQI for the three families in study 2 that completed intervention demonstrated improvements in the parents sleep quality following intervention, with global PSQI scores for all of the parents improving. Results of the PSQI suggest that all parents experienced improvements in their quality and efficiency of sleep. The ability to fall asleep, the quantity of sleep, and

the level of sleep disturbances either improved or remained unchanged for all parents. Of interest, is that Andrew had the most positive outcomes, with all scores either improving or remaining unchanged, despite Andrew not achieving all of his sleep goals. Previous research has found positive correlations between children with ASD and sleep problems, and poorer parental sleep quality (Lopez-Wagner, 2008; Meltzer, 2008. Meltzer & Mindell, 2007), but as these studies were cross-sectional, it was not possible to determine if changes to the children's sleep impacted on parents sleep quality. The current study is the first known study to demonstrate changes in parents sleep quality as a result of their child's sleep intervention.

RQI. Before intervention, parents of Matt, Catherine, Andrew, and Harry all had moderate to very high scores on the RQI, suggesting that they were all satisfied in their relationships. This is in contrast to previous research that suggests elevated levels of marital problems in parents of children with ASD (for example, Lopez-Wagner, 2008; Richdale, 2013). Results comparing pre-and post intervention scores on the RQI were reported for two families, with mixed results. Scores for both of Harry's parents remained high and consistent, whereas both of Andrew's parents reported decreases in their overall relationship satisfaction. It is possible that the sleeping intervention played a role in their RQI scores, but it is also possible that extraneous factors influenced these outcomes. The current study is the first known study to investigate the impact of children's sleep interventions on parents' relationship quality.

The current study shows, as suggested by previous literature, that sleep problems in children with ASD have far-reaching effects on the lives of their

families (Brown et al., 2013; Cortesi et al., 2010), and suggests that the treatment of a child's sleep problems can have collateral benefits for the lives of those around him (Tilford et al., 2015).

Parent understanding and acceptability of treatment. Overall, the findings of the current study suggest that for the four families that completed intervention, parents were satisfied with the intervention process. Another theme that was evident in the post-treatment interview was an appreciation for the daily contact with the researcher. Several participants said it created a feeling of support and allowed for immediate modifications to the intervention, resulting in faster and more effective results. Another theme was that the intervention was challenging to implement initially, but got easier as the intervention progressed.

These perceived outcomes were mirrored in the TARF-R results, with scores reflecting a high acceptance of the overall treatment process by all families. All families believed the treatment to be very reasonable and effective, they had a very good understanding of the treatment, and were willing to carry out the interventions. In contrast, families reported that the intervention required significant time and levels of disruption to the family, with some negative side effects. Interestingly, Andrew's mother reported a very high level of effectiveness despite not all of her sleep goals being met. Another point of interest is that Harry's parents reported the least amount of time/disruption as a result of the intervention, suggesting that standard extinction combined with a faded bedtime was the fastest and least disruptive intervention.

These high scores may be a result of the FBA process, as it provided parents with a valid rationale as to why particular treatments were chosen.

According to Weiskopp et al. (2005) FBA is an important means of aiding parental understanding of treatment method. Another benefit of FBA that was evident in this study was its ability to tailor interventions to fit within each child's unique family context and environment. Just as the factors maintaining sleep behaviours varied widely between children, so did the children's level of ability, their parents goals and motivations, levels of support and their home environment. These factors needed to be considered when designing intervention plans as interventions that fit better with the family are more likely to be implemented consistently, and adhered to, resulting in more successful outcomes (Moore, 2004). Collaboration with families throughout the treatment process was vital to ensure the interventions were achievable, socially valid, and comprehensible to each family, and adjusted to fit with the families observations, changing needs and preferences. This studies recognition of the importance of working with families to identify and treat sleep problems is in line with other studies that have emphasised the need to collaborate with families to achieve the most positive treatment outcomes (Jin et al., 2013; Moore, 2004; Turner & Johnson, 2013; Weiskop et al., 2005).

Limitations of the current study. There were several limitations to the current study, including the inability to isolate the effects of each treatment component, its reliance on psychometrics, difficulties generalising results, not obtaining inter-observer agreement for all participants, amount of therapist involvement, and lack of a long-term follow-up.

The experimental design of the current study means that changes in sleep behaviors can be attributed to the function of the changes in the sleep

environments that were a result of the intervention. However, one of the largest limitations of the current study is the inability to isolate the effects of individual treatment components. As all interventions were multi-modal, with components often delivered simultaneously, it is not possible to determine how each component contributed to the overall outcome, or whether effects were a result of the cumulative components. This is a limitation frequently cited by past researchers (for example, Knight & Johnson, 2014; Moore, 2004). While this is a limitation of this studies design, it is important to note that FBA often leads to multi-modal treatment packages, and therefore the current findings still have significant clinical implications.

Another limitation of the current study its the reliance on psychometrics to record changes in the child's daytime functioning and parents well-being as a result of the intervention. Psychometrics only identify changes in perceptions (Moore, 2004), and are therefore subject to a response bias as parents are not blind to the intervention (Weiskop et al., 2005). There is a need to quantitatively capture evidence of information provided anecdotally. In the instance of externalising daytime behaviour for example, frequency and duration recordings may more accurately reflect children's behaviour. Furthermore, it would be useful to detect any changes across different people and in different contexts (e.g., schools). The current study therefore serves as a preliminary study that highlights some of the most common areas subject to change as a result of a successful sleep intervention.

A third limitation of the current study is that due to the heterogeneity of factors maintaining presenting problems, and interventions, it may not be possible to generalise results to other children with ASD, even if they have similar

presenting problems. In an effort to combat this, participants were selected based on consistent presenting problems of co-sleeping and night wakings.

As video recordings were too intrusive for some families, this led to a failure to triangulate sleep related measures for Ben, Matt and Catherine, making IOA measures not possible for all children. This may threaten the validity of conclusions made for these children (Didden et al., 2002).

Another limitation was the amount of therapist involvement. In a clinical setting, daily contact may not be possible due to the cost and level of resourcing required. Therefore, the availability of support that families outside of this research might be provided with may not be reflected in the current study. However, in agreement with Moore (2004) and Weiskop et al. (2005), given the other significant stressors that families with children with ASD are likely to be experiencing, it is possible that this level of support for the parents who are the primary interventionists by default, is required for a sleep intervention to be successful. Both George's mother and Harry's father said that they benefited from the regular reinforcement and guidance that was provided by the researchers.

A final limitation is that long-term follow up data was not collected for Study 1 participants due to time constraints. It is intended that these will be carried out 12 weeks post intervention completion.

Recommendations for future research. The findings and limitations of the current study lead to several recommendations for future research. Firstly, a more thorough investigation into the impact of sleep interventions on parent wellbeing and child daytime functioning is warranted. The current study relied on psychometrics and anecdotal evidence to monitor changes in these areas. Both

areas could benefit from a more rigorous and comprehensive methodology where child behaviours are tracked and other aspects of family wellbeing are quantified before and after intervention.

Future research could investigate the impact of sleep interventions not only on parents, but on siblings as well. Ben's parents stated they believed his decrease in sleep disturbances had a positive impact on his brothers' sleep too, and it would have therefore been interesting to quantify this anecdotal information.

Several parents reported that partaking in a sleep intervention improved their self-efficacy and confidence around dealing with future sleep problems. Taking this into account, future research could measure parents' levels of self-efficacy pre- and post-intervention, and the extent to which their confidence extends to tackling other challenging behaviors.

Given the difficulty in isolating treatment effects, future research could determine what components of the treatment are most effective, and most acceptable to families. One way to investigate this, even when assessment informs the need for multiple treatments, would be to stagger the introduction of intervention methods so that the impact of each individual component could be assessed.

It is possible that the assessment process and interventions used in the study resulted in successful outcomes that are limited to the current population. Another recommendation for future research would be to expand the current study to include a wider variation of ages, capabilities, comorbidities, and developmental disabilities.

Research into the assessment of FBA to inform treatment for sleep disturbances in children with ASD, especially those with co-sleeping problems, which treatments are the most effective, and the impact of sleep interventions on daytime behaviors and parent well-being, is still very much in its infancy, and would benefit from further investigation.

Clinical implications. Interventions chosen in this study were based on interrupting and replacing factors that reinforced problematic sleep behaviors. The success of intervention lends support to a behavioural model of sleep disturbance (France & Blampied, 1993) common to both typically developing children and children with ASD. Despite all children in the study having similar presenting problems, different antecedent and consequence-based maintaining factors were identified for each individual, resulting in individualised methods. This highlights the importance of assessing sleep disturbance in children with ASD via a comprehensive process that identifies and addresses the specific factors maintaining the sleep problem for each individual. While FBA has been used extensively in the assessment of challenging behaviours, there are very few studies that have applied this process in the treatment of sleep difficulties in children (Jin et al., 2013). Some research has explored the effectiveness of antecedents or consequences in the treatment of sleep disturbances in the treatment of children with ASD, but very few have combined both antecedent and consequence variables to create comprehensive and individualised treatments based on FBA. This study can help to inform clinicians who are attempting to provide treatments for children with sleep-related problems.

The study found that improved sleep can have far-reaching effects on the child's behaviour and parents well-being. According to Cohen (2014), treatment guidelines to help manage challenging behaviors and individuals with ASD rarely recommend investigating sleep behaviors. This studies results imply that sleep in children with ASD should be considered when other behavioural challenges and/or parent's well-being is of concern.

While it is difficult to tease out the components of treatment that were responsible for changes in sleep for each individual, many practices were common across individuals and may be worth considering for use in clinical practice. This includes the use of visual supports and reinforcement. In addition, many treatment components were new tools to combat sleep problems in children with ASD, and should also be considered for use in clinical practice, for example stimulus substitutes and video modeling. Current findings also add to the existing evidence, supporting the use of extinction based procedures, and combined pharmacological and behaviour interventions to treat sleep problems.

Conclusion. The current study suggests that FBA is an important process to accurately inform sleep interventions for children with ASD. Multimodal behavioural treatments were able to eliminate the need for parent presence during sleep onset as well as co-sleeping during the night, and it was found to have a mostly positive effect on other sleep related measures. Successful sleep interventions were perceived to impact on the child's daytime functioning, especially externalizing behaviors, attention, and aggression, as well as overall ASD symptomatology. Successful sleep interventions also impact on parent well-being, with positive changes perceived in their own sleep quality. The impact of

intervention on parent's mental health and relationship satisfaction is less clear. In general, parents who completed intervention were satisfied with the intervention, had a good understanding of the treatment process, and believed it to be effective in improving their child's sleep behaviours. Overall, the study adds to the current literature around the use of FBA to inform interventions, the effectiveness of behavioral interventions for sleep problems in children with ASD, and the impact sleep interventions can have on the child's daytime functioning and parent well-being.

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Appendix A

Child Information Sheet



An investigation into the efficacy of non-traditional approaches to treat sleep disturbance in children with autism

Children's Information Sheet

Hello. My name is Laurie McLay and I am a lecturer at the University of Canterbury. I am doing a project about how to help children to sleep better and I would like for you to help me with this.

I am going to be talking to your parent/s about ways to help you to sleep better. This means that I will be coming to your house, or your parent/s will be coming to see me at the University.

I will ask you to wear a special watch called an actigraph sometimes. This will help me to understand the times that you are awake and asleep. There will also be a video camera in your bedroom sometimes. This will also help me to understand what you do when you are awake and asleep. Only your parents, and other people working on this project will be able to see this video.

If you do not want to be a part of this project, you can tell me or your parents and you won't need to be a part of it anymore.

If you have any questions you can ask me or your parents whenever you like.

Now we need to decide if you would like to do this. If you do want to be a part of my project then you can say "yes". If you do not want to be a part of this project then you can say "no" and no one will mind.

If you say yes, you or one of your parents can sign the form for you.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email human-ethics@canterbury.ac.nz

Appendix B
Child Consent Form



“An Investigation into the use of Functional Behaviour Assessment to Inform Intervention for Sleep Disturbance in Children with Autism Spectrum Disorders”

Children’s Consent Form

My name is _____.

Laurie has told me about the work that she is going to be doing with my parent/s.

Laurie told me that she is going to be working with my parent/s to help me to learn to sleep better.

I know that if I want to stop at any time or if I do not want to be a part of this project anymore, that will be fine. I can tell Laurie or my parents.

I was told that my parents/caregiver may sign this form for me and I think that is OK.

Child’s name: _____

Date: _____

Signature: _____

If this form is signed on behalf of your child please acknowledge, by signing this form, that your child was verbally informed of the investigation and what it will involve and that they were unable to provide verbal or written consent that they would like to be a part of this research.

Parent/caregiver: _____

Date: _____

Signature: _____

Please return this form to Laurie McLay.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email human-ethics@canterbury.ac.nz

Appendix C

Parent Information Sheet



An investigation into the efficacy of non-traditional approaches to treat sleep disturbance in children with autism or features of autism

Information for Parents/Caregivers

This research has been assessed and approved by the University of Canterbury Human Ethics Committee (HEC 2014/150).

Dear Parent,

We are a group of researchers at the University of Canterbury. Dr Laurie McLay is a lecturer in the School of Health Sciences at the University of Canterbury. Laurie has many years experience in working with children and young people with autism and their families. Associate Professor Karyn France has lectured here for many years, has conducted research into the treatment of paediatric sleep disturbance and is a registered clinical psychologist with considerable clinical experience in this area. Professor Neville Blampied has a similar history of teaching and research, and Jolene Hunter, Jacqui Knight, and Jenna van Deurs are students in Child and Family Psychology who will be working on their theses as part of this project.

We would like you to consider allowing your child with autism or Asperger's syndrome to participate in this research study. The primary purpose of this study is to investigate the effectiveness of treatments for sleep disturbance in children with autism or Asperger's syndrome. These treatment options include non-traditional approaches (e.g., massage therapy and white noise) as well as modified behavioural approaches. These approaches have been designed to minimise stress as much as possible for the parents and children using them. We are also interested in parents' experiences in using the treatments and any changes to their lives, or their child's lives, which result.

If you agree to allow your child to be a part of this study, we will meet with you to discuss your child's sleep behaviour with you and find out more about him/her and your family. This initial meeting will last for approximately 1 hour. We will then ask you to complete sleep diaries in which you will record further information about your child's sleep patterns. Sleep diaries will be recorded each day throughout all phases of the study as this will allow us to monitor the effectiveness of the treatment approach. The sleep diaries will take you up to five minutes to complete each night. You will also be asked to complete a commonly used questionnaire in order to obtain information about your child's sleep behaviour and the effects of treatment. It will take approximately 15 minutes to complete this questionnaire. When we have established an understanding of your child's sleep behaviour, we will work with you to develop sleep-related goals for your child. This will involve a second treatment planning meeting which will last 1-1 ½ hours.

To help us gather further information about your child's sleep patterns we will bring a video camera to your home for some nights over the course of the

programme, that is capable of recording all night sleep. This method will allow us to measure sleep behaviour at times when an adult is not present. We will demonstrate and explain how to use the video equipment for gathering information.

As a part of this study we would also like to investigate the experiences of families in implementing treatments for sleep disturbance, those treatments that they consider to be most acceptable, and the impact of successful treatment of sleep problems on parent and child wellbeing and quality of life. In order to do this we will ask you to complete some questionnaires about your and your child's well-being and behaviour at the commencement and conclusion of treatment. We will also ask your perspective on the treatment that was provided. We will do this either during visits to your home or in a clinic at the University of Canterbury.

When information about your child's sleep behaviour has been gathered, treatment will commence. You will be offered a choice of treatment options. The treatment will be implemented for up to four weeks. If you are dissatisfied with the treatment approach or the degree of progress that is being made then you will be offered a choice of another non-traditional approach, or alternatively, a modified behavioural approach to treatment can be implemented. If you would prefer to use a behavioural approach from the beginning then this is also an option. We will provide you with all of the necessary information about each treatment approach and we will maintain regular contact with you during treatment. It is anticipated that your involvement in the study will be over a period of 3-4 months.

Your child will be assigned a code name to ensure anonymity and anything that you or your child says or does will be kept confidential. The results of the study may be submitted for publication to national or international journals and may also be presented at conferences.

If you want to withdraw from the project before completion, you can do this at any time without penalty or repercussions.

Should you require any additional information about the study or if you would like to access the study findings you are able to do so at any stage. The data which is produced from the research will be kept in a locked cabinet at the University of Canterbury for a minimum of ten years.

If you agree for your child to take part in the research, please sign the consent form that is attached.

If you have any complaints you may contact the Chair of the University of Canterbury Ethics Committee. The contact details are given below.

If you have any questions about this project please feel free to contact me:

Phone: 64 (3) 364-2987 ext. 7176

Email: laurie.mclay@canterbury.ac.nz

Appendix D

Parent Consent Form



CONSENT FORM FOR PARENTS

This research has been assessed and approved by the University of Canterbury Human Ethics Committee.

I wish to participate in the project, “An Investigation into the use of Functional Behaviour Assessment to Inform Intervention for Sleep Disturbance in Children with Autism Spectrum Disorders”.

I have read and understood the information that was given to me about this study.

I understand what will be required of me and my child/the child in my care during this project

I understand that the investigators do not foresee any potential risks to me or my child as a result of participating in this study.

I understand that all information about my family will be treated as confidential unless there is concern about anyone’s safety. In this case my clinician will need to speak to someone else to ensure the safety risk is removed. No findings that could identify me or my child will be published.

I understand that the findings of this study may be published in a research journal or at a conference and that the anonymity of me and my child will be maintained.

I understand that participation in this project is voluntary and that I can withdraw my child or he/she can withdraw from the project at any time without repercussions. I can also withdraw any data that has been collected at any time prior to the publication of that data.

I understand that all research data that is collected will be securely stored at the University of Canterbury for a minimum of ten years.

I understand that I am able to request a copy of the results of this research, should I wish to do so, and that these results will be provided for me.

I allow video-taping of my child’s sleep behaviour to be completed by the researcher and understand that this videotape will be used for data gathering purposes only. I also understand that I have the right to request that video footage is destroyed at any stage.

I consent to others, listed below, being involved in the implementation of the intervention.

Name: _____

Date: _____

Signature: _____

Others I consent to implementing intervention:

Name: _____

Name: _____

Name: _____

Please return this form to Laurie McLay.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email humanethics@canterbury.ac.nz

Appendix E
Audio-visual Consent Form



Pukemanu – Dovedale Centre
Child and Family Psychology Services
University of Canterbury

“An Investigation into the use of Functional Behaviour Assessment to Inform Intervention for Sleep Disturbance in Children with Autism Spectrum Disorders”

AUDIO-VISUAL RECORDING CONSENT FORM

You have been given this form because the researcher and/or student/intern Psychologist has asked your permission to make an audio-visual recording of his/her session with you. Reviewing sessions with supervisors and other students is an important part of psychologist training and ensuring research integrity.

Please read the statements below, which explain the purpose of audio-visual recording and how your privacy would be protected:

- The purpose of recording this session is to:
 1. Enable the student/intern Psychologist and her supervisor(s) to review, evaluate and discuss the student/intern Psychologist's performance.
 2. Gather data for the research project.
- Audio-visual recording of any session will only be done with your knowledge and consent.
- You can withdraw your consent to a session being recorded and/or replayed, either before the session commences, or after it has been recorded, without having to provide a reason for changing your mind.
- The audio-visual file will only be seen by the researchers and student/intern Psychologist's.
- The audio-visual recording will be securely stored at the University of Canterbury for a minimum of ten years.
- I understand that I have the right to request that the tape be turned off at any point during the sessions.

I hereby consent to have an audio-visual recording made on the above conditions.

Your Signature:

Researchers/Student's Signature:

Date:

Appendix F

Initial Screening

Begins with an explanation about confidentiality (*see consent forms in appendix B & D*).

Screening questions used (over the telephone);

- 1) Tell me what led up to you enquiring about the study. (How did you hear about it, where you referred by someone? –details)
- 2) How old is your child? /What is your child's date of birth?
- 3) The programme is for children with autism and limited verbal ability. Tell me about your child in relation to that:
 - What is your child's diagnosis? Tell me about getting the diagnosis- who diagnosed your child.
 - Do they have any secondary diagnoses?
 - Does your child have any physical or medical conditions that may contribute toward their sleep difficulties?
 - Is your child on any medication?
- 4) Tell me about your child's sleep patterns?
- 5) What are your primary concerns with regard to this sleep behaviour?
- 6) Is your child currently receiving any services or treatment for their behaviour, including sleep?
- 7) The next step is for us to meet at the University so I can get full information about your child and the problem. Do you have any questions at this point?

Appendix G Standard Sleep Diary Template

	Date:	Monday __	Tuesday	Wednesday	Thursday __	Friday __	Saturday _	Sunday __
Daytime sleep	Setting							
	Time put to bed							
	Time awake							
Night-time sleep	Setting							
	Time put to bed							
	Frequency of Curtain calls							
	Curtain calls after put to bed (Code and describe each)	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:
	Your responses to each (Code and describe each)	I R F T L Other:	I R F T L Other:	I R F T L Other:	I R F T L Other:	I R F T L Other:	I R F T L Other:	I R F T L Other:
	Time until silence							

Appendix G continued Standard Sleep Diary Template

		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
Nigh time awakenings	Time & Duration of awakening	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	
	Behaviour while awake (Code)	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:	Q S V B P C Other:
	Your response (Code)	I R F T L Other:	I R F T L Other:	I R F T L Other:	I R F T L Other:	I R F T L Other:	I R F T L Other:	I R F T L
	Woke at...	//////////	//////////	//////////			//////////	//////////

Record a code for the behaviours that were observed when your child was put to bed and while awake. Also record a code for your response to these behaviours.

Behaviour codes: **Your child** - Squealing (Q); Self-Stimulating (S); Vocalising (V); Bouncing (B); Playing with toys/activities (P); Crying (C); Bowel motion (BM), leaves room (LR)

Your Response – Ignored (I); Physically returned to bed (R); Offered food or drink (F); Provided with activity or toys (T); Lay with them (L)

If behaviour isn't covered by one of these codes, then please write down the behaviour that was observed.

Notes

Appendix H

Example of Clinical Interview Questions

Begin with confidentiality terms and obtain consent to be interviewed;

Problem behaviour

- Tell me about what your concerns are?
- Walk me through a typical night?
- Problem behaviour; frequency, duration, setting, when does it occur, when does it not occur, what happens immediately after the behaviour, what makes it better/worse, what do you do when the behaviour occurs?
- Opinion- what do you think causes the behaviour?
- Sleep hygiene- what is the current bed time routine, time and place of bed, wake up time

History of problem behaviour

- Approximately how long has the behaviour been occurring?
- Have there been changes in the frequency or intensity of behaviour over time?
- What attempts have been made in the past to change the behaviour? Were they successful?

Developmental History

- How was the pregnancy, birth, parent reactions to having a child at the time?
- What was his behavioural style like as an infant?
- Did he meet all his milestones?
- Who are the significant people in his life?
- Do you have any concerns for him, aside from sleep?

Families of origin

- How do you think your own backgrounds have impacted on your parenting style?
- What supports do you have in place at the moment?

Summary and termination

Appendix I

SATT Example Questions (Jin et al., 2013)

Sleep Problem and History

Please provide a description of your child's sleep problems

How long have these problems occurred?

Sleep Goals

Describe your goals regarding your child's sleep

Labelling Specific Sleep Problems

Does your child have difficulty going into the bedroom at night when instructed to do so or brought in to go to sleep?

If yes, his behavior pattern is referred to as **night-time routine noncompliance**

Please provide some details

If yes, what do you usually do to help your child to go to bed?

If yes, what usually works to get your child to go to bed?

Once in bed, does your child have difficulty staying in bed or remaining still in bed when instructed to do so?

Does your child repeatedly call out or engage in other behavior that requires you to return to his/her bedroom?

If yes, this behavior pattern is referred to as **interfering behavior**

Please provide some details

If yes, what do you do to help your child to stay in bed?

If yes, what usually works to get your child to stay in bed?

If yes, what usually works to get your child to stop the "call outs?"

Once in bed, does your child have difficulty falling asleep (i.e., it typically takes more than 15 min for her to fall asleep)?

If yes, his behavior pattern is referred to as **delayed sleep onset**

Please provide some details

Sleep Schedule

At what time does your child typically go to bed?

At what time does your child typically fall asleep?

At what time does your child wake in the morning?

How many hours does your child sleep at night?

How many hours does your child sleep during the day?

At what time does your child typically go down for a nap?

At what time does your child typically wake from a nap?

Appendix J

Outline of Post-Treatment Discussion

Purpose: informal interview/discussion to gauge parent's perspective of the treatment process, and hear their thoughts and feedback.

- How do you make sense of the improvement?
- What is it that you both did, that you feel made a difference?
- How did you find the intervention overall, and the process?
- Did the child's progress/improvement have an impact on you personally, if so- how?
- Did you notice any other changes in your child's behaviour in response to intervention?
- Any suggestions for how process could have been improved?
- Any questions?

Appendix K
George's parental presence and Vallergran dosage regime

Day	Vallergran (ml)	Mother's actions
1	1.8	Move (end of bed)
2	1.8	
3	1.8	Move (chair next to bed)
4	1.4	
5	1.4	
6	1.4	Move (chair further from bed)
7	1.1	
8	1.1	
9	1.1	Move (chair further from bed)
10	0.8	
11	0.8	
12	0.8	Move (chair next to door)
13	0.5	
14	0.5	
15	0.5	Move (chair out of door)
16	0.2	
17	0.2	
18	0.2	